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Long-term dyadic adjustment of parents of children with acute lymphoblastic leukemia: Couples' experiences from treatment completion to survivorship

par Willow Burns

Département de psychologie
Faculté des arts et sciences

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Résumé

Les taux de survie chez les patients pédiatriques atteints de leucémie lymphoblastique aiguë (LLA) se sont progressivement améliorés avec le temps, de sorte que la grande majorité des enfants diagnostiqués avec cette maladie chronique survivent. Bien que ce soit une nouvelle encourageante pour le patient et sa famille, être diagnostiqué et traité pour cette maladie dans l'enfance nécessite encore un ajustement considérable de la part de l'enfant malade et de la famille, en particulier chez les parents. À la suite du diagnostic de leur enfant, les parents prennent souvent des responsabilités supplémentaires pour s'occuper de l'enfant malade et ces responsabilités vont au-delà de leurs responsabilités parentales préexistantes. De telles exigences peuvent être très lourdes pour les parents au plan individuel et relationnel. Plusieurs études et revues empiriques ont porté sur l'adaptation psychologique des parents au cancer pédiatrique, mais seulement récemment, l'accent a été mis sur l'impact du cancer pédiatrique sur les relations des parents. Bien que ces études aient été fondamentales pour approfondir notre connaissance du fonctionnement des couples, elles ont principalement utilisé des approches individuelles pour leur analyse et ont été axées sur les expériences d'ajustement à court terme, peu après le diagnostic ou alors que l'enfant est encore en traitement. Il y a encore peu de données sur l'ajustement à long terme des couples. Le principal but de cette thèse est de proposer une approche dyadique pour comprendre les expériences psychologiques et d'ajustement relationnelle à long terme des parents d'enfants atteints de leucémie lymphoblastique aiguë (LLA).

Le premier article a servi de test initial de l'approche dyadique sélectionnée, *le modèle d'interdépendance acteur-partenaire*, avec une cohorte longitudinale de parents dont l'enfant a

été diagnostiqué et traité pour une leucémie lymphoblastique aiguë (LLA). En particulier, cet article visait à déterminer si l'ajustement conjugal des partenaires deux ans après le diagnostic de leur enfant pouvait être prédit par les états d'humeur perçus par les partenaires et leur perception du fonctionnement de leur famille au moment du diagnostic. Quarante-sept couples ont rempli des questionnaires d'auto-évaluation pour évaluer ces domaines au moment du diagnostic (états d'humeur des parents, le fonctionnement familial et l'ajustement conjugal initial) et deux ans plus tard (ajustement conjugal). L'ajustement conjugal des mères à la fin des traitements LLA de leurs enfants (deux ans après le diagnostic) a été associé à leur propre perception du soutien familial, des conflits de rôle et de la surcharge de rôle au moment du diagnostic. L'ajustement conjugal des pères à la fin du traitement a été associé à leur propre conflit de rôle, à leur ambigüité de rôle et à leur fatigue au moment du diagnostic, ainsi qu'à la perception par les partenaires du conflit de rôle au moment du diagnostic.

S'appuyant sur les résultats et l'expérience de cette première étude dyadique et en étendant notre analyse à la période de survie, notre deuxième article visait à examiner les associations entre l'ajustement à long terme des couples et les souvenirs de la dynamique de relation passée avec leur partenaire. Une cohorte de suivi de 103 couples d'enfants ALL survivants a été invitée à réfléchir au moment où son enfant a été traité et à se rappeler la nature des changements de relation (changement négatif, sans changement, changement positif) sur des dimensions relationnelles spécifiques, comme l'intimité ou la sexualité. Ils ont également été invités à décrire leur bien-être psychologique actuel et leur fonctionnement en relation. Les résultats montrent que les partenaires au sein des couples ont eu tendance à être en accord sur la nature des changements dans les relations qui se sont produits suivant le diagnostic de leur enfant. Les mères décrivent qu'une meilleure satisfaction relationnelle

actuelle a été associée à leur propre perception des changements positifs de la relation, alors que la meilleure relation relationnelle des pères était associée à la perception par les partenaires des changements de relation positifs. La détresse psychologique actuelle des mères était associée à leur propre perception des changements de relation et la détresse psychologique actuelle des pères était associée à la fois à leur propre *et* aux perceptions de leurs partenaires sur les changements de relation. Ces résultats suggèrent que l'ajustement psychologique et relationnel des mères de l'enfant avec leucémie est un processus individuel lorsque l'ajustement des pères est un processus surtout interdépendant, soutenant ainsi la nécessité d'utiliser une approche dyadique pour comprendre l'ajustement de ces couples. Afin de favoriser l'ajustement des deux partenaires, les cliniciens devraient se concentrer sur les besoins de chaque partenaire, en particulier ceux des mères.

Mots-clés: parents, couples, leucémie pédiatrique, détresse, relation, ajustement, dyade, post-traitement, survie

Abstract

Survival rates for pediatric patients with acute lymphoblastic leukemia (ALL) have gradually improved over time, with the vast majority of children diagnosed with this chronic disease surviving. Although this is encouraging news for the patient and his family, being diagnosed and treated for this disease in childhood still requires considerable adjustment on the part of the sick child and the family, especially for the parents. Following their child's diagnosis, parents often take on additional responsibilities to care for the sick child and these responsibilities go beyond their pre-existing parental responsibilities. Such demands can be very cumbersome for parents at individual and relational levels. Several empirical studies and reviews have focused on the psychological adjustment of parents to pediatric cancer, but only recently has the emphasis been placed on the impact of pediatric cancer on parents' relationships. Although these studies were fundamental in deepening our understanding of couples' functioning, they mainly used individual approaches for their analysis and focused on short-term adjustment experiences either shortly after diagnosis or when the child is still being treated. There is still little data on the long-term adjustment of couples. The main goal of this thesis is to propose a dyadic approach to understand the long-term psychological and relational adjustment experiences of parents of children with acute lymphoblastic leukemia (ALL).

The first article served as an initial test of the selected dyadic approach, *the Actor-Partner Interdependence Model*, with a longitudinal cohort of parents whose child was diagnosed and treated for ALL. In particular, this article examined whether the marital adjustment of partners two years after their child's diagnosis could be predicted by partners'

perceived mood states and their perception of family functioning at the time of diagnosis. Forty-seven couples completed self-assessment questionnaires to assess these areas at the time of diagnosis (parental moods, family functioning and initial marital adjustment) and two years later (marital adjustment). Mothers' marital adjustment at the end of their child's ALL treatments (two years after diagnosis) was associated with their own perceptions of family support, role conflicts and role overload at the time of diagnosis. Fathers' marital adjustment at the end of treatment was associated with their own role conflict, role ambiguity and fatigue at the time of diagnosis, as well as the partners' perception of role conflict at the time of the diagnosis.

Based on the results and experiences of this first study dyadic and extending our analysis to the survival period, our second article examined the associations between long-term adjustment of couples and memories of past relationship dynamics with their partner. A follow-up cohort of 103 couple of childhood ALL survivors were asked to think back to when her child was treated and remember the nature of the relationship changes (negative change, no change, positive change) occurring on specific relational dimensions, such as intimacy or sexuality. They were also asked to describe their current psychological well-being and relationship functioning. The results show that the partners within couples tend to agree on the nature of relationship changes that occurred after their child's diagnosis. Mothers' describing stronger current relationship satisfaction was associated with their own perception of positive changes in their relationship, whereas fathers' stronger interpersonal relationship was associated with their partner's perception of positive relationship changes. Mothers' current psychological distress was associated with their own perception of relationship changes and fathers' current psychological distress was associated with both their own *and* their partner's

perceptions of relationship changes. These results suggest that the psychological and relational adjustment of mothers of children with ALL is an individual process, while the adjustment of fathers is a mostly interdependent process, hence supporting the necessity of using a dyadic approach to understand these couples' adjustment. In order to foster both partners' adjustment, clinicians should focus on addressing each partner's needs, especially those of mothers.

Keywords: parents, couples, pediatric leukemia, distress, relationship, adjustment, dyad, post treatment, survivorship

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List of abbreviations

ALL: Acute lymphoblastic leukemia

APIM: Actor-Partner Interdependence Model

MAT: Locke-Wallace Marital Adjustment Test

FWA: Family Well-Being Assessment

POMS-BI: Profile of Mood States - Bipolar Form

DAS-4: Dyadic Adjustment Scale - 4 items

BSI-18: Brief Symptom Inventory - 18 items

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Introduction

Diagnosis of pediatric leukemia is a psychologically and interpersonally stressful hardship that can significantly disrupt and challenge not only the life of the ill child, but also the family at large, especially the parents (Long & Marsland, 2011). Parents are expected to be a pillar of strength and stability for the family, and when a child is diagnosed with a chronic illness, such as leukemia, this source of strength and stability is even more sought after. Following the child's cancer diagnosis, parents become primary caregivers and partner with their child's health care team in the aim of providing the most comprehensive and supportive environment possible to foster their child's recovery (Hutchinson, Willard, Hardy, & Bonner, 2009). For this reason, it is essential that parents' take care of themselves both in terms of their psychological well-being and their relationship well-being as a couple. Both facets of their well-being are important, as they can help parents provide the support and resilience that the ill child and family need from them. It is equally important that parents provide mutual support to one another in the context of their relationship as a couple. Also, according to a longitudinal study of parents of children with cancer, both partners can significantly influence each other's marital adjustment (Dahlquist, Czyzewski, & Jones, 1996), thereby suggesting that couples' adjustment is interdependent. Findings regarding these interrelationships in couples' adjustment were also reported by another longitudinal study of parents of pediatric cancer patients (Hoekstra-Weebers, Jaspers, Kamps, & Klip, 1998). Nevertheless, most studies on couples' adjustment in relation to pediatric cancer have used a cross-sectional design and individual approaches to understand adjustment (da

Silva, Jacob, & Nascimento, 2010; Van Schoors, Caes, Alderfer, Goubert, & Verhofstadt, 2016). Commonly, research in this field has examined parents' and couples' adjustment using the same select time points within the child's cancer trajectory: diagnosis and during treatment. Thus, emphasis has been placed primarily on their short-term adjustment. Significant less empirical focus has been placed on understanding their long-term adjustment, as assessed following treatment completion and in the survivorship period (e.g., Ljungman et al., 2014). Use of different time points is beneficial in comparing how parents and couples adjust over time, thus capturing their adjustment as a process not a snapshot of one moment during the illness. Therefore, parents' roles as influential primary caregivers and the possible interrelationships between partners' adjustment, led us to the aims of the present thesis.

The present thesis provides a dyadic approach for understanding the long-term psychological and relationship adjustment experiences of parents of children with ALL, by considering the experiences of both partners in the couple. The introduction that follows provides an overview of the empirical literature in this field including its limitations, which served as both a rationale and catalyst for the research program of this thesis. Besides the theoretical background, the introduction also presents the conceptual model and dyadic approach that were used in this thesis, as well as its underlying research objectives. Following the introduction, the two research articles included in this thesis are presented as two consecutive chapters. The final chapter acts as a general discussion of the findings from these two respective studies, as well as its potential clinical and theoretical implications.

Pediatric ALL: Illness trajectory and challenges

According to the Canadian Cancer Society (2017), 32% of all new cases of childhood cancer in Canada are leukemia, thus signifying the largest incidence of all childhood cancer types. Leukemia can appear in different forms. The thesis at hand will focus on ALL, the most commonly diagnosed leukemia among children representing 75 to 80 percent of all childhood leukemias (Dana-Faber Cancer Institute, 2017). ALL is a cancer that affects both the bone marrow and the white blood cells. The most common symptoms of childhood ALL are: fever, bruising or petechiae (small red dots on the skin indicating low platelet counts), anemia (low red blood cell count), dyspnea (difficulty breathing), bone and joint pains, abdominal pain, swollen lymph nodes, and recurring infections (Dana-Faber Cancer Institute, 2017). When a child is diagnosed with ALL, treatment commences shortly thereafter and it lasts approximately 2 to 3 years (American Cancer Society, 2017a). Treatment for pediatric ALL is primarily chemotherapy (American Cancer Society, 2017a), however in cases where the child is at high risk of relapse radiation therapy might also be necessary. In rare cases, stem cell transplants (or bone marrow transplants) from donors are used to replace stem cells that were damaged by intensive treatments with chemotherapy or radiation therapy (Dana-Faber Cancer Institute, 2017). The standard progression of the child's treatment for ALL includes three phases, with each having their own particularities and stresses for parents (American Cancer Society, 2017a).

Given that the present thesis focuses on parents' adjustment to their child's ALL diagnosis and treatments, it is essential to understand the medical context of this pediatric malignancy. By understanding the particularities of each illness stage, we will be better

able to understand the associated psychological and relationship challenges faced by parents of children with ALL, both on an individual and dyadic basis. The first phase following diagnosis is *induction*, and this phase lasts typically approximately 1 month (American Cancer Society, 2017a). This phase represent the initial crisis of having one's child diagnosed with a chronic illness. Accordingly, this time can be very demanding for parents as they are confronted with adjusting to this new reality and their caregiving responsibilities. During induction, their child can experience prolonged hospitalizations for treatment, severe infections (which can be fatal), or other complications (American Cancer Society, 2017a). Chemotherapy drugs (e.g., asparaginase, vincristine) and corticosteroid drugs (e.g., prednisone, dexamethasone) are given to pediatric patients right away during this first phase of treatment (American Cancer Society, 2017a; Cooper & Brown, 2015). For children that are at a high risk of ALL relapse, their chemotherapy can be more intensive than for other standard risk patients, or they can be given radiation therapy in addition (American Cancer Society, 2017a). The goal is to use a first block of chemotherapy to attain *remission* by the end of this phase (American Cancer Society, 2017a; Cooper & Brown, 2015)). Reaching remission does not mean that the child is completely cured from their cancer. Instead, it implies that most leukemia cells are no longer present in the bone marrow, normal cells are reappearing, and blood counts are starting to be normal (American Cancer Society, 2017a). An astounding 95% of pediatric ALL patients reach remission by the end of induction (American Cancer Society, 2017a; Cooper & Brown, 2015). However, it is possible that pediatric ALL patients achieve remission in other treatment phases instead.

Next, the child enters the *consolidation* (or *intensification*) phase of treatment, a 1- to 2-month phase characterized as being even more intense than the preceding phase (American Cancer Society, 2017a). The goal is to continue destroying remaining leukemia cells (American Cancer Society, 2017a). Combinations of chemotherapy drugs are given to the child to help fight these leukemia cells and to prevent the body from developing drug resistance (American Cancer Society, 2017a). At this time in the cancer trajectory, some high-risk patients undergo more extended or intensive consolidation treatments (Cooper & Brown, 2015), or stem cell transplants (American Cancer Society, 2017a). Parents might find this time particularly taxing as they witness their child enduring intense treatment regimes that are physically demanding and almost always include side effects due to toxicity (e.g., nausea or vomiting) (Cooper & Brown, 2015)

The final treatment phase is *maintenance*, and this phase is the longest in duration, spanning roughly 2 years in duration (American Cancer Society, 2017a). In order to start maintenance treatment, the child has to still be in remission after the end of the induction and consolidation phases (American Cancer Society, 2017a). Typically this treatment phase has less toxicity and is offered on a largely outpatient basis (Cooper & Brown, 2015). Both chemotherapy drugs and steroids are administered to the patient. Within the first few months of maintenance, there are intensified 4-week treatment periods that resemble the induction phase. These periods are referred to as *re-induction* (American Cancer Society, 2017a). Although this phase, occurring roughly 2-year post diagnosis, might at first glance appear to have less pressing concerns for parents than the earlier two phases, it is an influential time within the illness trajectory. The child's treatments are concluding, and this time can be conceptualized as a transition phase whereby parents

might be experiencing heightened stress in relation to the end of their child's treatment (Cincotta, 1993). Although the end of treatment is a significant milestone, parents are faced with new challenges. For instance, as the child's primary caregivers, parents are required to closely monitor their child's health and support their child with less involvement from the health care team. One parent in a mixed methods study on the long-term psychosocial effects of pediatric cancer, qualitatively described this experience at the end of treatment as: *"When you bring your child home, that's when you're totally alone"* (Quin, 2005, p. 141). Parents are also faced with the challenge of adapting to a new reality post cancer, including addressing their own anxieties that were repressed during treatment, increasing child discipline regarding illness-related limitations, and fearing the possibility of cancer relapse (Muskat et al., 2017; Quin, 2005).

According to a recent review by Ljungman et al. (2014), a *childhood cancer survivor* (CCS) can be defined as an individual diagnosed with cancer as a child who is currently at least 5 years post diagnosis or at least 2 years post treatment. Survival rates for childhood ALL have progressively improved over time (Hunger et al., 2012). In 1990 to 1994 survival rates for ALL in children (who were less than 15 years old) were 80.2%, however by 2000 to 2004 these rates had increased to 87.5% (Hunger et al., 2012). Now, most children diagnosed with ALL become long-term survivors, with 85% of them becoming event-free survivors and 90% being completely cured of the illness (Dana-Faber Cancer Institute, 2017). Although children with ALL are surviving longer, the long-term and late effects of their illness are still significant concerns for many of these survivors and their parents (Iwai et al., 2017). The toxicity and intensity of childhood leukemia treatments can lead to various physical and psychosocial long-term and late

effects. Recent reports conducted in the context of either the Childhood Cancer Survivorship Study (CCSS) or the St Jude Lifetime Cohort Study suggest that childhood cancer survivors often experience issues with: psychological distress (e.g., anxiety, depression; (Brinkman et al., 2016; Schultz et al., 2007), fertility (Green et al., 2009; Green et al., 2017; Wasilewski-Masker et al., 2014), inattention or hyperactivity (Brinkman et al., 2016; Jacola et al., 2016), learning (Jacola et al., 2016), social withdrawal or antisocial behaviour (Brinkman et al., 2016; Schultz et al., 2007), and cardiac insufficiencies such as congestive heart failure or myocardial infarction (Mulrooney et al., 2009). Also, a St Jude Lifetime Cohort Study report on adverse health outcomes among 1713 adult survivors of childhood cancer, found that the most prevalent issues were impairments in: pulmonary (65.2%), auditory (62.1%), endocrine or reproductive (62.0%), cardiac (56.4%), and neurocognitive (48.0%) functions (Hudson et al., 2013). Research has also insisted on the impact of cancer on the social environment surrounding the ill child, including their family and friends. Cancer relapse can occur at any given time (Dana-Faber Cancer Institute, 2017), likewise fear of recurrence has been commonly cited as a significant concern for parents of childhood cancer survivors (Ljungman et al., 2014). Parents of childhood cancer survivors also frequently report psychological distress (e.g., anxiety, depression, posttraumatic stress symptoms) and physiological complaints (e.g., sleep disturbances and fatigue, somatic symptoms) (Ljungman et al., 2014). In turn, these worries and symptoms can potentially spill over into other aspects of their life, such as in their relationship with their partner.

Parents as primary caregivers: Adjusting to a new reality

The child's cancer diagnosis is a life-changing event for the entire family and typically serves as a catalyst for substantial changes in family dynamics (da Silva et al., 2010; Long & Marsland, 2011; Vrijmoet-Wiersma et al., 2008). While there are several stakeholders in the family that can be influenced by the child's cancer, parents' involvement particularly stands out. Given that ALL is typically diagnosed in children that are between 3-to 5-years of age (St. Jude's Children's Research Hospital, 2017), this active involvement is particularly understandable. As parents of a chronically ill child, they are solicited to work with their child's health care team and take on new caregiving and illness management responsibilities. In this way, parents' become caregivers that are integrally involved in their child's illness trajectory (Hutchinson et al., 2009). A cross-sectional study conducted by Hutchinson et al. (2009) compared the psychological adjustment of caregivers of children with brain tumours that are undergoing active treatment and caregivers of children with brain tumours that are off-treatment. They found that while off-treatment caregivers reported significantly less general distress, both caregiver groups reported significant caregiving burden and uncertainty regarding their child's illness (Hutchinson et al., 2009). This finding appears to suggest that caring for a child with cancer is a significant and enduring burden for parents throughout the entire illness trajectory. It can be especially demanding and stressful for parents to try to juggle all these new responsibilities with their pre-existing parental responsibilities (Long & Marsland, 2011; Vrijmoet-Wiersma et al., 2008). As conceptualized by McCubbin, Balling, Possin, Frierdich, and Bryne (2002), this division of family responsibilities has been referred to as "living as a split family," with one parent being the primary caregiver

for the ill child in the hospital and the other parent being responsible for all other family responsibilities (e.g., household chores, caring for the other children, maintaining employment; Long & Marsland, 2011; McGrath, 2001). This split-family dynamic typically endorses traditional gender roles in the division of family responsibilities (Clarke, McCarthy, Downie, Ashley, & Anderson, 2009). Mothers tend to stay with the ill child at the hospital, whereas fathers tend to be responsible for the household and financial responsibilities of the family (Clarke et al., 2009; Long & Marsland, 2011). In qualitative interviews, fathers have described feeling overwhelmed with balancing all these responsibilities and conflicted in their role (Chesler & Parry, 2001). In particular, being distant from the ill child and mother can leave fathers feeling isolated and excluded from important treatment decisions (Chesler & Parry, 2001; McGrath, 2001). Their desire to be present for their partner and ill child at the hospital is at odds with their desire to care for the other children at home (McGrath, 2001). In turn, this can lead fathers to report feeling considerable gender role conflict (Chesler & Parry, 2001; Hall, 2010a; McGrath, 2001), and difficulty maintaining their view of themselves as the provider and protector of their family (Chesler & Parry, 2001). Likewise, a quantitative survey on communication, social support, and gender role conflict among parents of children found that when fathers experienced more gender-role conflict (i.e., career achievement conflict concerning his personal worth as a financial provider) they were less anxious, but they perceived getting less support (instrumental and emotional) from their partner. Mothers traditionally leave paid employment (sometimes permanently) to be with the ill child at the hospital. In fact, a Canadian pilot study found that 64% of mothers left their jobs after diagnosis compared to 16% of fathers (Limburg, Shaw, & McBride, 2008). Parents'

likelihood of leaving their jobs was higher if the child was less than 10 years old at the time of diagnosis and if the diagnosis was leukemia (Limburg et al., 2008). Mothers have also been more disadvantaged in terms of their income after their child's cancer diagnosis. Compared to controls, both mothers and fathers of children with cancer report income reductions in the year of their child's diagnosis. However, this reduction is significantly greater for mothers (21% reduction for mothers versus 10% reduction for fathers; Lindahl Norberg, Montgomery, Bottai, Heyman, & Hovén, 2017). Additionally, it takes mothers of children with cancer 6 years to reach the income level of control mothers while for fathers catching up only takes 3 years (Lindahl Norberg et al., 2017).

Gender differences. Parental distress can vary as a function of time since diagnosis. Actually, distress of both mothers and fathers of children with cancer is most pronounced near the time of diagnosis, with mothers typically reporting significantly more distress than fathers. Several reviews have reported significant gender differences in parents' distress and adjustment at the time of diagnosis or when the child was undergoing active cancer treatments (Pai et al., 2007; Sultan, Leclair, Rondeau, Burns, & Abate, 2015; Vrijmoet-Wiersma et al., 2008). During treatment, mothers consistently report significantly greater psychosocial distress than fathers, irrespective of type of distress (global distress, anxiety, depression, somatization, posttraumatic stress symptoms; Vrijmoet-Wiersma et al., 2008). In fact, according to a review by Pai et al. (2007), mothers report more distress than fathers up to one year following diagnosis. However, mothers do not simply report more distress than fathers, but more of them also meet clinically significant thresholds for their distress (Vrijmoet-Wiersma et al., 2008). Once the child's treatment is completed, the distress levels of mothers and fathers are

said to be comparable (Maurice- Stam, Oort, Last, & Grootenhuis, 2008). During the survivorship period, most parents' psychological distress has subsided considerably, however a small subgroup of parents still report clinically significant distress, with 21 to 44% of parents reporting severe posttraumatic stress symptoms five or more years following their child's cancer diagnosis (Ljungman et al., 2014).

Couples' adjustment

Individual adjustment is simply referring to an individual's assessment of psychological functioning, whereas couples' adjustment is referring to the relationship functioning of the couple. Couples' adjustment includes such global predictors of relationship well-being as: relationship satisfaction, marital distress, and relationship adjustment. Compared to parents' individual adjustment to pediatric cancer (Barrera, Atenafu, Doyle, Berlin-Romalis, & Hancock, 2012; Hutchinson et al., 2009; Sultan et al., 2015; Vrijmoet-Wiersma et al., 2008), couples' adjustment is a relatively new field of study within the pediatric cancer literature (da Silva et al., 2010; Lavee & Mey-Dan, 2003; Long & Marsland, 2011). To date, quantitative and qualitative studies alike have reported rather mixed findings on couples' adjustment. While some couples reported that their child's cancer had a primarily negative impact on their relationship (e.g., deteriorations in sexuality), others report that it also had a positive impact (e.g., improvements in trust, communication, partner support, and emotional closeness; Brody & Simmons, 2007; da Silva et al., 2010; Lavee & Mey-Dan, 2003; Van Schoors et al., 2016; Wiener et al., 2016). Studies on couples' adjustment have examined global constructs of relationship functioning such as relationship satisfaction or marital adjustment from the point of view of one of the partners, most often this being the mother

(Van Schoors et al., 2016), but only a minority of studies have recruited both partners to described their relationship functioning.

In general, couples' relationship functioning appears to fluctuates in a curvilinear fashion over the course of the child's illness trajectory (Lavee, 2005; Lavee & Mey-Dan, 2003). The child's cancer diagnosis marks the beginning of the illness trajectory, and in a cross-sectional study of 134 parents of children recently diagnosed with cancer roughly 25% of mothers and 28% of fathers reported clinically significant marital distress (Dahlquist & Czyzewski, 1993). Another smaller scale cross-sectional study with 35 couples found that couples' retrospectively recalled that their relationship satisfaction decreased somewhat in the first year following their child's diagnosis (Lavee & Mey-Dan, 2003). This finding was further paralleled by a longitudinal study on parents of pediatric cancer patients, which found that parents' marital dissatisfaction increased significantly in this first year post diagnosis (Hoekstra-Weebers et al., 1998). As demonstrated by the above studies, the first year following their child's cancer diagnosis is undoubtedly a very taxing period on the parents' relationship as a couple. If the child has been ill for two or three years, couples recalled that there was a slight increase in their relationship satisfaction. However, in cases when the child has been ill for four or more years, couples' relationship satisfaction declined again (Lavee & Mey-Dan, 2003). Thus, the most pronounced deteriorations in the couples' relationship occur during the first year of the child's illness (i.e., the initial crisis) and in cases where the illness was enduring (lasting four or more years) (Lavee & Mey-Dan, 2003).

Thus far, only two cross-sectional studies have attempted to further dissect the specific aspects of the couples' relationship that have changed following their child's

cancer diagnosis beyond global concepts such as relationship satisfaction or marital adjustment (Lavee & Mey-Dan, 2003; Wiener et al., 2016). The first of these two studies was retrospective in nature and smaller in scope as it only investigated the experiences of 35 couples that had a child diagnosed with cancer (Lavee & Mey-Dan, 2003). Yet, its findings helped identify the relationship dimensions that significantly changed following their child's cancer diagnosis, as well as couples' agreement on the nature of these relationship changes (positive change, negative change, or no change). Overall, these couples tended to report improvements in communication and trust with their partner, and marked deteriorations in sexuality (Lavee & Mey-Dan, 2003). They perceived no significant change in their conflict resolution, leisure activities and division of household responsibilities. High agreement between partners was reported on: conflict resolution, leisure activities, sexuality, relationship with extended family, and interpersonal trust (Lavee & Mey-Dan, 2003). The second cross-sectional study aimed at disentangling specific relationship effects was much larger in scope, representing the experiences of 192 parents (122 mothers and 70 fathers) of children currently on treatment or in follow-up care (Wiener et al., 2016). According to this study, a third of parents reported low dyadic adjustment and that their relationship quality had deteriorated. In fact, following the child's cancer diagnosis more than half of these parents stated that their relationship with their partner had been challenged (Wiener et al., 2016). Also, specific aspects which men and women found to be particularly stressful were also examined in this study. These findings suggest that partners differed on a few core concerns. For instance, men were more likely than women to indicate that lacking intimacy with their partner was particularly stressful for them (Wiener et al., 2016).

Select factors are particularly influential in differentiating couples that report positive relationship changes from those that report negative changes. These associated factors include: shorter illness duration, greater social support for mothers, and greater sense of coherence for fathers (i.e., belief that they can cope with stress satisfactorily; da Silva et al., 2010; Lavee, 2005). A few other core constructs have been associated with parents' adjustment as a couple, namely emotional distress and self-rated coping styles. Using a cross-sectional design, Dahlquist and Czyzewski (1993) found that general distress (which included both anxiety and depression), the difference between parents' state anxiety levels, and their use of sensitization coping (i.e., tendency to approach a stressful stimuli as opposed to avoiding it) were significant predictors of marital distress for both mothers and fathers. Findings from a subsequent study conducted by Dahlquist et al. (1996) extends their scope from a cross-sectional design to a longitudinal one. In this longitudinal study of 42 couples of children with cancer, mothers' marital adjustment at treatment follow-up (20-months post diagnosis) was predicted by their self-reported depression and fathers' marital satisfaction near diagnosis (2 months post diagnosis). Fathers' marital adjustment at follow-up was predicted by their child's health status, their self-reported depression, and mothers' marital satisfaction near diagnosis (Dahlquist et al., 1996). Specifically, their child's poor health status contributed to greater marital adjustment at follow-up, and these researchers hypothesized that for fathers their child's deteriorating health could be associated with their reliance on their marriage for support (Dahlquist et al., 1996). Another longitudinal study echoes the interrelated nature of partners' marital adjustment. On the one hand, mothers' marital distress at 6 and 12 months post diagnosis was associated with their partner's coping preferences (problem-

focused or emotion-focused). On the other hand, fathers' marital distress was associated with their own self-reported coping preferences (Hoekstra-Weebers et al., 1998).

Limitations and gaps in the literature

Despite the theoretical and practical merit of studies on couples' functioning in relation to pediatric cancer, most of these studies share notable limitations. To begin with, studies that examine couples' adjustment or functioning following their child's cancer diagnosis have almost exclusively been conducted using individual approaches. Hence, these studies are not actually investigating couples' adjustment, but instead reflect each parent's individual reports of relationship adjustment. Since the couple is not the unit of analysis, adjustment reports might only be reflecting individual well-being. Another fundamental issue with using individual approaches for understanding couples' variables is that they are not considering the possibility that one partner is influencing the other, thereby suggesting interrelationships in couples' adjustment.

Although more studies in recent years have included both mothers and fathers of children with cancer in their examination of relationship functioning (Khoury, Huijer, & Doumit, 2013; Lavee, 2005; Lavee & Mey-Dan, 2003; Wijnberg-Williams, Van de Wiel, Kamps, & Hoekstra-Weebers, 2015), there are still a plenitude of studies that only focus on the experiences of one parent (Brody & Simmons, 2007; McGrath & Chesler, 2004; Tremolada et al., 2013; Young, Dixon-Woods, Findlay, & Heney, 2002). Indeed, a systematic review on couples' functioning after their child's cancer diagnosis indicated that 35% of the studies included only one partner from the couple (Van Schoors et al., 2016). In some cases this might be due to convenience sampling (i.e., recruiting the parent that is more readily available), while in other cases the researcher might purposely

recruit only one parent due to the aims of their study. Although, even when both parents' participation is solicited, the final sample might still end up representing the experiences of one parent more than the other. Mothers are typically over represented in pediatric cancer samples in comparison to fathers. Actually, a recent review on the impact of childhood cancer on parents' relationship confirmed this exact phenomenon, with 63.5% of participants being the mother of the ill child (Wiener et al., 2016).

Numerous past studies on couples' functioning in pediatric cancer settings are also limited by their reliance on cross-sectional designs. Likewise, a review study on couples' functioning conducted by Van Schoors et al. (2016) reported that roughly two thirds of the studies included in their review were cross-sectional. Cross-sectional designs are correlational in nature, and cannot discern process issues. Only longitudinal designs can assess temporal associations involved in couples' adjustment to pediatric leukemia. This proposition regarding the temporality of adjustment is supported by findings from another review on the impact of pediatric cancer on couples' relationships. Specifically, da Silva et al. (2010) reported that time since diagnosis or illness duration is the most common clinical characteristic related to couples' adjustment in pediatric cancer settings.

In addition, less empirical emphasis has been placed on parents' long-term distress and adjustment following treatment completion and in the survivorship phase than in all other illness stages. A review on the survivorship period assessed parents' adjustment primarily as their long-term individual psychological distress (posttraumatic stress symptoms, anxiety, fear of cancer reoccurrence; Ljungman et al., 2014). Similarly, in a qualitative study on the experiences of parents' five years after the completion of their child's cancer treatments or death, parents described still being anxious especially

when reflecting on their child's illness and the possibility of relapse. They also described that negative feelings which they suppressed during their child's treatments were more intense now that treatment was completed (Ljungman et al., 2016). Future studies on parents of childhood cancer survivors should specifically evaluate both facets of long-term adjustment: psychological and relational.

Lastly, most studies on couples' functioning in the context of pediatric cancer are atheoretical, in that they lack a conceptual model to support their research propositions. Use of a guiding theory is beneficial, especially in terms of facilitating early interventions. By having an in-depth understanding of the adjustment process, health practitioners and counsellors will be better able to identify couples that are struggling and provide them with additional support. Indeed, knowing what maladaptive relationship dynamics could be undermining couples' adjustment, in turn helps determine specific targets for improvement in couples' therapy. Future studies in this field should: 1) include both partners within the couple, 2) conceptualize adjustment as including both its psychological and relational components, and 3) use a dyadic approach to understand both partners' adjustment experiences and the ways in which one partner might be influencing the other partner's adjustment (i.e., interrelationships).

Conceptualizations of individual and dyadic stress

Traditionally research has viewed stress individually as reflecting either: 1) a *stimulus* (e.g., an important life event) that elicits stress reactions (Dohrenwend, 1974), 2) a *reaction* to a significant demand (Selye, 1974) or 3) a *process* involving both the person and their environment (i.e., transactional view of stress; Lazarus & Folkman, 1984). However, recently there has been a shift towards also interpreting stress as being

dyadic in nature (Bodenmann, 2005; Randall & Bodenmann, 2009). In fact within couples' research, stress and adjustment are both key concepts. Given that stress and conjugal distress can be related to one another (Randall & Bodenmann, 2009), how couples' adjust to stressful life events can significantly impact both partners' well-being. According to Bodenmann (2005), *dyadic stress* is a specific form of stress within a social system (e.g., couples' relationship, marriage, or family). It can be conceptualized as *"a stressful event or encounter that always concerns both partners, either directly or indirectly"* (Bodenmann, 2005, p. 34). This definition implies that the stressor is of mutual concern, meaning that both partners will need to appraise the stress and take measures to cope both individually and as a couple (Bodenmann, 2005). Specifically, the couples' shared or common efforts to address or deal with the source of stress are referred to as *dyadic coping* (Bodenmann, 2005). This demonstrates the conceptual link between dyadic stress and dyadic coping. Dyadic coping includes three core elements: common concerns regarding the source of stress, joint goals in addressing the stress, and interdependence between the partners. Accordingly, dyadic coping can have a dual impact, in that it can contribute to partners' stress reduction and their relationship enhancement (Bodenmann, 2005). Individual coping and dyadic coping should not be seen as mutually exclusive events. Actually, it is very likely that both individual and dyadic coping are occurring simultaneously when a couple and the partners within that couple are confronted with a shared stress (Bodenmann, 2005).

Pediatric leukemia as a dyadic stress

A recent review by Randall and Bodenmann (2017) proposed that stress can be seen as being a dyadic experience for couples, with both partners being able to

reciprocally influence one another. A chronic illness can be conceptualized as one such source of stress for couples (Randall & Bodenmann, 2017). Likewise, the illness can be viewed a dyadic or interpersonal phenomenon requiring dyadic coping by both partners (Badr & Acitelli, 2005; Badr & Acitelli, 2017; Berg & Upchurch, 2007; Fife, Weaver, Cook, & Stump, 2013; Randall & Bodenmann, 2017). Past research has conceptualized cancer as a "we-disease,' given that it can affect both the adult patient, as well as their partner (Kayser, Watson, & Andrade, 2007).

Recent research on couples in pediatric cancer settings have started to explore this sense of we-ness (or integration of the couples' relationship into self-identity) and its impact on conjugal resilience (Martin et al., 2014; Martin, Péloquin, Vachon, Duval, & Sultan, 2016). Pediatric leukemia is embedded in two influential social systems, the family and parents as a couple. Typically, parents are the primary caregivers for the ill child and the rest of the family, thus their individual and dyadic well-being is of paramount importance. Diagnosis of pediatric leukemia is a shared concern for both parents and it can have a significant and enduring impact on both partners in the couple. Likewise, both partners will need to implement coping strategies to face this hardship. These coping strategies need to address their common concerns as a couple (e.g., fear of treatment complications or relapse) and their shared goals (e.g., becoming a unified team in the face of adversity). Given these considerations, pediatric leukemia can and should be regarded as a dyadic stress for the couple. Furthermore, since pediatric leukemia is a chronic illness, adjustment of pediatric patients and their parents might be best conceptualized as a temporal process, beginning at the time of diagnosis and continuing thereafter. This implies that adjustment is not a static process, and that different stages in

the illness trajectory may have characteristic challenges associated with fluctuations in couples' adjustment over time.

Dyadic model of couples' adjustment

Provided that pediatric leukemia has been conceptualized as a dyadic stress, it is fitting that the unit of analysis for the present thesis be the dyad. A dyad implies that two individuals are connected in some way (Kenny, Kashy, & Cook, 2006), and in the case of this thesis that connecting element is their role as parents to the ill child. It is probably entirely unrepresentative to examine both parents' adjustment independently since they are confronted with the same dyadic stress (i.e., having their child diagnosed and treated for ALL). Instead of examining each partner's experiences individually, dyadic models allow us to investigate both partners' experiences at the same time (Kenny et al., 2006). They also allow us to examine associations between partners' experiences (i.e., interrelationships). In fact by using a dyadic model, we directly account for the interdependent nature of couples' adjustment (Kenny et al., 2006).

One such dyadic model is the *Actor-Partner Interdependence Model* (APIM), and this was the selected dyadic model used in the present thesis (APIM; Kenny et al., 2006). This particular dyadic model has three core advantages when compared to individual approaches: 1) it assumes the interdependence of couples' data, 2) it statistically tests actor and partner effects simultaneously, and 3) it statistically tests for gender differences in these effects (Kenny et al., 2006). An *actor effect* represents the association between a partner's predictor and his or her own respective outcome, whereas a *partner effect* assesses the association between a partner's predictor and their partner's outcome (Kenny et al., 2006). Such a dyadic model might better encapsulate the complex and intricate

nature of parents' adjustment in pediatric leukemia settings. APIM models have been used in studies with couples in which one partner has cancer (e.g., breast cancer, prostate cancer, lung cancer; Götze et al., 2017; Lee, Kim, Lim, & Kim, 2016; Manne et al., 2015; Schellekens et al., 2017). In the context of pediatric cancer, only two other research teams worldwide (Compas et al., 2015; Hall, 2010a; Hall, 2010b) have published reports thus far using the Actor-Partner Interdependence Model. Using APIM models with a sample of 150 couples, Compas et al. (2015) found that mothers' disengagement coping strategies were associated with fathers' greater depressive symptoms, even after controlling for fathers' own coping strategies and sociodemographic variables. In a smaller study with 22 couples, Hall (2010a) found that fathers' effective communication was related to mothers' perceiving greater social support, and in turn this was related to mothers reporting less anxiety. Another APIM-based study by Hall (2010b), suggested that during their child's cancer treatments both mother and fathers receive quality support from shared sources of social support (e.g., the parent of one particular spouse), however when it comes to unique sources of social support, mother received significantly greater high quality instrumental support than fathers. Together these three cross-sectional studies advocate for the merit and use of this dyadic model for assessing inter-partner effects among parents of children with cancer.

Conceptual model and thesis objectives

The framework underlying this thesis is the *Resiliency Model of Family Stress, Adjustment and Adaptation* (McCubbin et al., 2002), which has previously been applied to pediatric cancer by Brody and Simmons (2007). This resiliency model is composed of: the stressor event (A), the family's resources or abilities to adapt to the stressor event (B),

the meaning that the family has given to the stressor event (C), and the extent of crisis experienced by the family (X). In the context of the present thesis, the focus was on the couple not on the family at large, so the Resiliency Model was adapted to reflect this specification (refer to adapted figure below). The stressor event (A) was the child's ALL diagnosis, and adjustment was conceptualized as the couples' adjustment. Taken together, all our adjustment outcomes were psychosocial in nature, reflecting the psychological and social adjustment of both parents. This Resiliency Model served as an inspiration for the two studies conducted within this thesis. Each of the above components were assessed, however all components were not assessed in the same study. This was due to our desire to conserve the particularities of a few select illness stages (diagnosis, end of treatment, and survivorship period), and allow for greater specification into couples' adjustment at these moments within their child's illness trajectory. Thus, we opted to only conserve components that were hypothesized as being specifically influential at the given time of assessment. As such, we selected two separate paths in this Resiliency Model: the ABX path and the CX path.

The two studies included in the present thesis shared the overarching aims of addressing core limitations in the emergent field of couples' relationships in pediatric cancer settings by: 1) investigating clinical and social factors that are associated with the long-term adjustment of both mothers and fathers of children with ALL in two large samples of couples; and 2) identify interrelationships between partners' adjustment by considering the couple as the unit of analysis. We also decided to address a critical gap in the empirical literature by investigated couples' long-term adjustment both nearing the

end of treatment (2-years post diagnosis) and in the survivorship period (5 or more years post diagnosis).

Study 1. When referring to the Resiliency Model, we selected the ABX path to be the focus for the first study of the present thesis. The stressor event (A) was the child's ALL diagnosis. Emphasis was placed on understanding the associations between parents' resources (B) at the time of diagnosis (perceptions of family well-being and mood states) and their marital adjustment (X) nearing the end of their child's ALL treatments (i.e., 2-years post diagnosis). This study served as an initial test of the actor-partner interdependence model in the context of pediatric leukemia. Specifically, it aimed to: 1) determine whether family functioning and parental mood in both partners near the time of their child's ALL diagnosis were associated with both partners' marital adjustment 2 years following diagnosis, and 2) determine whether there were significant gender differences in these effects. Using a longitudinal, dyadic design, this study allowed us to assess each partner's predictors of marital adjustment over time, as well as the possibility that adjustment was interdependent (with one partner's perceptions at diagnosis being associated with the other partner's adjustment 2-years post diagnosis). Examining couples' adjustment at this time is essential for the implementation of early interventions aimed at helping partners adjust to their dual roles as parents and caregivers. This first study (presented as Chapter 3) was published in *Psycho-Oncology*.

Study 2. In reference to the Resiliency Model, path CX was selected as the emphasis for the second study of this thesis. The aim of this second study was to examine both partners' adjustment (psychological and relationship) in the survivorship period, and determine whether partners' adjustment was related to their own or their partner's reports

of relationship changes that were recalled as having occurred during their child's ALL treatment. This study addressed an apparent gap in the empirical literature, as couples' relationship adjustment has not previously been investigated in the survivorship period. This study also addressed a core limitation of the first study, which is the failure to examine the ways in which the couple's relationship has changed following their child's leukemia diagnosis. This second study took this into account and precisely examined changes in various relationship dimensions perceived by both partners during treatment. It also expanded on the first study by examining parents' long-term adjustment as including both psychological distress and relationship satisfaction. The multifaceted nature of adjustment was an inherent advantage to this second study. In this retrospective, cross-sectional study, emphasis was placed on the meaning that was assigned to the stressor event (i.e., relationship changes that each partner perceived) and each partner's adjustment (i.e., psychological distress and relationship satisfaction) once their child has entered the survivorship period. The objectives of this study on parents of childhood ALL survivors were to: 1) describe couples' long-term adjustment; 2) describe the perceived impact of cancer on the couples' relationship; and 3) identify to what extent the perceived impact of cancer on the couple was related to both parents' long-term psychological and relationship adjustment. A novel feature of this study was that it directly examined relationship dimensions (e.g., intimacy, sexuality) and the nature of changes each partner recalled experiencing on these relationship dimensions following their child's leukemia diagnosis (i.e., negative impact, no impact, or positive impact on these relationship aspects). In this way, it could allow for greater specification on which aspects of couples' relationship tend to be strengthened and those that tend to be hindered as a result of the

child's illness, and their respective impact on current psychological and relationship functioning. By understanding aspects that are strengthened and that favourably impact couples' adjustment later, we are identifying pillars of strength. Couples should be encouraged to use their strengths in the face of adversity, and early interventions targeting areas of difficulty for couples could also promote better long-term adjustment. This second study (presented as Chapter 4) was submitted for publication in *PLoS ONE*.

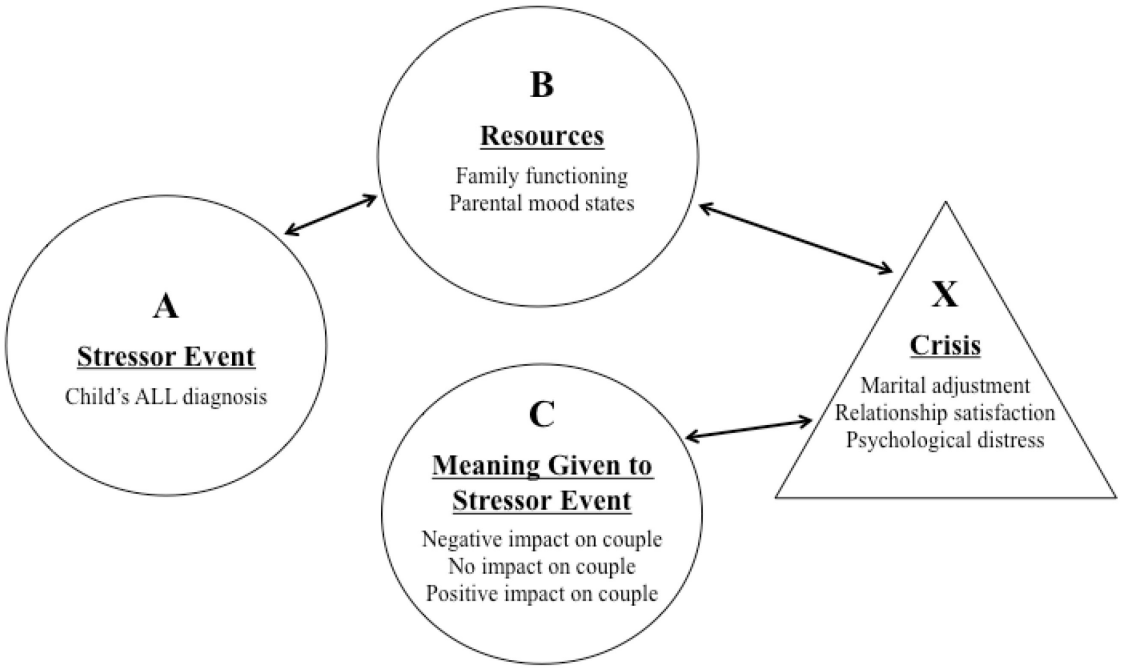

Figure 1. *Adapted Resiliency Model (Adapted from Brody & Simmons, 2007)*

Methodological Approach

The current doctoral research program consisted of the examination of two related but separate databases. Both Study 1 and 2 were based on Quebec cohorts of parents of children with acute lymphoblastic leukemia, who were diagnosed and treated at the

Sainte-Justine University Health Centre. These two studies were conducted exclusively in French. The two studies were additive in nature, meaning that each one builds upon the former. Both studies used the same dyadic statistical approach, *the actor-partner interdependence model*. However, there was a noteworthy methodological difference between these studies. Specifically, Study 1 was longitudinal in design, and Study 2 was retrospective and cross sectional in nature.

Study 1. Study 1, presented as Chapter 2 in the current thesis, was an exploratory secondary analysis on a pre-existing database (Qc ALL 91-95; PI: Dr. Caroline Laverdière). The underlying aim of Study 1 was to provide an initial test of the application of the actor-partner interdependence model in the clinical context of couples whose child was treated for leukemia. Earlier findings from the Qc ALL 91-95 database, that discuss the neurodevelopmental impact of ALL treatments and behavioral problems experienced by these patients have been published elsewhere (Marcoux, et al., 2013; Marcoux, Robaey, Krajinovic, Moghrabi, & Laverdiere, 2012; Robaey, Krajinovic, Marcoux, Moghrabi, 2008). Children (18 years or less) diagnosed and treated for acute lymphoblastic leukemia (ALL) at the Sainte-Justine University Health Centre between February 1993 and September 1999, and their parents were recruited to participant in the initial study (138 families) (Marcoux, et al., 2013; Marcoux et al., 2012). These pediatric ALL patients were treated with either DFCI protocols 91-01 or 95-01. This initial longitudinal study followed these children and parents from the time of diagnosis up to 4 years post diagnosis, with self-report measures being completed by parents individually at diagnosis, 3-months post diagnosis, 1-year post diagnosis, 2-years post diagnosis, 3-years post diagnosis, and 4-years post diagnosis. For Study 1, we only focused on data

from two clinically relevant time points, at the time of diagnosis (i.e., the initial crisis) and 2-years post diagnosis (i.e., at or nearing the end of the child's ALL treatment). Only parents of these children who were in a couple with the other parent of the ill child at both the time of diagnosis and 2-years later were retained for secondary analysis.

Study 2. Study 2, presented as Chapter 3 in this thesis, was also an exploratory study using the same dyadic analysis approach as Study 1. Participants for Study 2 were recruited through the project "Prévenir les effets des traitements à long terme dans la leucémie lymphoblastique aigue" (i.e., PETALE project). Thus, Study 2 was conducted in the context of this larger multidisciplinary CIHR funded study (PI: Dr. Daniel Sinnett). The PETALE project was a cross-sectional description of long-term effects of ALL, which were retrospectively linked to clinical history.

Parents whose children were part of the Qc ALL cohort that were treated at Sainte-Justine University Health Centre following DFCI protocols were contacted if they met eligibility criteria for participation in the PETALE project. This project included childhood ALL survivors who were 19 years old or less at the time of diagnosis, and were diagnosed and treated at the Sainte-Justine UHC with DFCI protocols since 1989. To participate in the PETALE project, children also needed to be under 19 years of age, not have experienced relapse, and be at least 5 years post diagnosis at the time of the PETALE study. These ALL survivors were included in a comprehensive medical examination of their metabolic, cardiac, neurological, and psychoaffective health. Eligible parents were asked to complete a series of questionnaires when their child came for their yearly visit at the long-term clinic. If one parent was not present to complete the questionnaires at this time, a stamped envelope containing the questionnaires was given

to the parent that was present or to the child in cases of parental separation. These stamped envelopes could be mailed to the researcher coordinator upon completion.

There was some overlap in samples between Study 1 and 2, namely Study 1 represented approximately 30% of the whole PETALE cohort. The main component of the PETALE project, which was used in the context of this doctoral program consisted of asking parents of ALL survivors to reflect on their relationship as a couple and how it was impacted by their child's illness. This sub-cohort of children and their parents served as the participants for Study 2.

Co-Authors' Contributions

The author of this thesis, Willow Burns, took an active role in preparing, conducting, and disseminating the research of both studies included within this thesis. For the first article, presented as Chapter 2, the data was initially collected in the context of Dr. Sophie Marcoux' doctoral thesis, which was co-supervised by Dr. Philippe Robaey and Dr. Maja Krajinovic. Upon receiving permission from them to perform secondary analyses on their database, the author of the thesis conceptualized the aim of the exploratory study, selected the methodological approach, prepared and analyzed the data, and wrote the manuscript. This research was co-supervised by Dr. Serge Sultan and Dr. Katherine Péloquin. All co-authors contributed their input before publication of the manuscript. For the second article, presented as Chapter 3, project administration and data collection for the PETALE study, which included Study 2 of this thesis, was done by a team of researcher coordinators and assistants (Émélie Rondeau, Simon Drouin, Laurence Bertout, and Ariane Lacoste-Julien). The author of the thesis was responsible for the following tasks: conceptualization of the study and its methodology, data

preparation and analysis, and writing the manuscript. The author of the thesis also acquired a doctoral fellowship from the Cole Foundation and support from the CHU Sainte-Justine Foundation for this research. Additional research funding for the psychosocial part of the PETALE study was obtained by: Dr. Maja Krajinovic, Dr. Caroline Laverdière, Dr. Daniel Sinnett, and Dr. Serge Sultan (FRQs 29206 PI Sultan). This research was co-supervised by Dr. Serge Sultan and Dr. Katherine Péloquin. All co-authors contributed their input before publication of the manuscript.

Article 1

A 2-year dyadic longitudinal study of mothers' and fathers' marital adjustment when caring for a child with cancer

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TITLE:

A 2-year dyadic longitudinal study of mothers' and fathers' marital adjustment when caring for a child with cancer

AUTHORS:

Willow Burns, MA (PhD Candidate), Department of Psychology, University of Montreal, Montreal, Quebec, Canada; Research Center, Sainte-Justine University Health Center, Montreal, Quebec, Canada.
Katherine Péloquin, PhD, Department of Psychology, University of Montreal, Montreal, Quebec, Canada.
Serge Sultan, PhD, Department of Psychology & Pediatrics, University of Montreal, Montreal, Quebec, Canada; Research Center, Sainte-Justine University Health Center, Montreal, Quebec, Canada.
Moghrabi, Albert, MD, Research Center, Sainte-Justine University Health Center, Montreal, Quebec, Canada.
Sophie Marcoux, PhD, Department of Pediatrics, University of Montreal, Montreal, Quebec, Canada; Research Center, Sainte-Justine University Health Center, Montreal, Quebec, Canada.
Maja Krajinovic, MD, Department of Pediatrics, University of Montreal, Montreal, Quebec, Canada; Research Center, Sainte-Justine University Health Center, Montreal, Quebec, Canada.
Daniel Sinnett, PhD, Department of Pediatrics, University of Montreal, Montreal, Quebec, Canada; Research Center, Sainte-Justine University Health Center, Montreal, Quebec, Canada.
Caroline Laverdière, MD, Department of Pediatrics, University of Montreal, Montreal, Quebec, Canada; Research Center, Sainte-Justine University Health Center, Montreal, Quebec, Canada.
Philippe Robaey, MD, PhD, Research Center, Sainte-Justine University Health Center, Montreal, Quebec, Canada; Research Institute, Children's Hospital of Eastern Ontario, Ottawa, Ontario. Canada; Department of Psychiatry & School of Psychology, University of Ottawa, Ottawa, Ontario, Canada.

CONTACT INFORMATION OF CORRESPONDING AUTHOR:

Serge Sultan, PhD
Research Center, Sainte-Justine University Health Center
3175, Chemin de la Côte-Sainte-Catherine
Montreal, Quebec, Canada, H3T 1C5
Tel. + 1 (514) 343-6111 (20727)
Email : serge.sultan@umontreal.ca

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Abstract

Objective: Studies examining interrelationships within parental couples confronted with pediatric cancer are scarce. This study explored dyadic longitudinal associations between both partners' family functioning and mood at diagnosis, and marital adjustment two years later.

Method: Parents of children (*n*=47 couples) with acute lymphoblastic leukemia (ALL) completed the Family Well-Being Assessment and Profile of Mood States-Bipolar Form at diagnosis, and the Locke-Wallace Marital Adjustment Test two years post diagnosis. Multilevel linear models using the actor-partner interdependence model (APIM) and controlling for baseline marital adjustment were conducted to evaluate within subject and dyadic longitudinal effects.

Results: For mothers, better marital adjustment two years post diagnosis was associated with perception of greater family support and less role conflict and role overload at diagnosis. For fathers, better marital adjustment two years post-diagnosis was associated with perception of less role conflict, greater role ambiguity, and being more tired at diagnosis, as well as their partner's perception of less role conflict at diagnosis.

Conclusions: These findings highlight the importance of considering both partners' perspectives in understanding marital adjustment across treatment phases in parents of children with ALL. Early interventions for couples should be tailored to meet each partner's needs in order to foster resilience within the couple.

Keywords: pediatric cancer; parents; oncology; marital adjustment; family functioning; APIM

Introduction

The diagnosis of a pediatric cancer is undoubtedly a difficult time for parents. The demands associated with the child's illness and treatment can lead to changes in family dynamics, roles, and responsibilities (Vrijmoet-Wiersma et al., 2008), and may also bring challenges for the marital relationship (Long & Marsland, 2011). While confronted with the threat of their child's death, parents are called to make crucial and difficult decisions about their ill child's treatment and care. Parents' reactions to diagnosis and their ability to readjust their roles effectively in the face of cancer will likely influence both their own adjustment and their child's adjustment (Long & Marsland, 2011; Sultan, Leclair, Rondeau, Burns, & Abate, 2015). Although the effects of pediatric cancer on parents' individual functioning have been widely cited (Barrera, Atenafu, Doyle, Berlin-Romalis, & Hancock, 2012; Hutchinson, Willard, Hardy, & Bonner, 2009; Sultan et al., 2015; Vrijmoet-Wiersma et al., 2008), research on the impact of cancer on their conjugal functioning has received limited attention (da Silva, Jacob, & Nascimento, 2010; Lavee & Mey-Dan, 2003; Long & Marsland, 2011). This research also generally has important limitations, including the use of cross-sectional designs and reliance on using an individual perspective to explain marital outcomes instead of formal dyadic approaches considering both partners' perspectives. Dyadic studies examining predictors of long-term marital adjustment in the context of pediatric cancer are thus needed. The parental couple is a fundamental component within the family system that could potentially serve a protective function for both the ill child and the family. Understanding the factors associated with better relationship adjustment over time therefore appears crucial.

The handful of studies that have examined the impact of pediatric cancer on marital functioning have yielded mixed results. Quantitative and qualitative studies found that while some couples report a negative impact of their child's illness on their relationship, others report relatively little change or a positive impact, including improved support, trust, and communication (Brody & Simmons, 2007; Lavee & Mey-Dan, 2003; Shapiro, Perez, & Warden, 1998). However, this dichotomous classification into positive and negative relationship changes is likely not representative of the intricate and evolving nature of the cancer experience. Caring for a child with cancer can affect parents' life as a couple and the quality of the relationship may follow a temporal adaptation process, reflecting particularities of stages in the cancer trajectory. Past cross-sectional studies suggest that marital quality follows a curvilinear course as a function of illness duration (Lavee, 2005; Lavee & Mey-Dan, 2003). Near the time of diagnosis, parents tend to report heightened marital dissatisfaction and distress (Dahlquist & Czyzewski, 1993; Dahlquist, Czyzewski, & Jones, 1996; Yeh, 2002). Research found that among couples whose child was newly diagnosed, 25% of mothers and 28% of fathers had clinically significant marital distress scores (Dahlquist & Czyzewski, 1993). This initial marital distress and dissatisfaction would endure throughout the first year following diagnosis, as indicated by both cross-sectional (Lavee & Mey-Dan, 2003) and longitudinal studies (Dahlquist et al., 1996; Hoekstra-Weebers, Jaspers, Kamps, & Klip, 1998). Another cross-sectional study found that when the child had been ill for two to three years, couples tended to report slightly less marital dissatisfaction, but if the child's illness had progressed for four or more years, marital dissatisfaction tended to increase

again (Lavee & Mey-Dan, 2003). These observations thus justify the importance of longitudinal studies examining relationship adjustment.

Research suggests that what differentiates couples who report positive relationship changes versus deteriorations following a diagnosis of pediatric cancer are: shorter illness duration, mothers' having greater social support, and fathers' belief that they can cope with stress effectively (Lavee, 2005). Psychological distress or mood disturbances were also associated with greater marital distress for both partners two months following diagnosis (Dahlquist & Czyzewski, 1993), and partners' relationship distress 20-months post diagnosis was predicted by their own depression and their partner's marital dissatisfaction at diagnosis (Dahlquist et al., 1996). Marital distress predictors for mothers and fathers may also differ. Whereas mothers' marital distress at 6 and 12-months post diagnosis was associated with her partner's coping at 6 and 12-months (i.e., other-related), fathers' was associated with his own psychological distress and coping near diagnosis (i.e., self-related) (Hoekstra-Weebers et al., 1998).

Although the pediatric oncology literature has identified some predictors for marital adjustment, most studies were cross-sectional and used an individual approach. Longitudinal and dyadic studies are scarce and although dyadic studies have been reported in adult cancer settings (e.g., (Lafaye et al., 2014; Manne & Badr, 2010; Moser, Künzler, Nussbeck, Bargetzi, & Znoj, 2013)), only two dyadic studies have been conducted in the pediatric cancer setting (Compas et al., 2015; Hall, 2010a). In a sample of 22 couples, the first study found, among other results, that a parent's effective communication was related to their partner's perception of greater social support, and perception of greater social support was related to lower anxiety in mothers (Hall,

2010a). In a sample of 150 couples, the second dyadic study found that mothers' disengagement coping strategies were related to fathers' greater depressive symptoms, after controlling for fathers' own coping strategies and sociodemographic variables (Compas et al., 2015). Findings from these studies underscore the pertinence of assessing inter-partner effects to enhance our systemic understanding of both parents' adjustment in the context of pediatric cancer. Both of these studies used a cross-sectional design and did not examine marital adjustment, and thus cannot discern how both partner's initial perceptions and adjustment to the illness contribute to their relationship functioning over time.

In the current dyadic longitudinal study, we aimed to determine whether family functioning and parental mood in both partners near the time of diagnosis would predict both partners' marital adjustment 2-years post diagnosis. The Circumplex Model of Marital and Family Systems (Olson, Russell, & Sprenkle, 1983) proposes that two dimensions of family interactions are relevant to understand adaptation. The model has been applied in the context of families of chronically ill children (Kazak, 1989). *Cohesion* refers to families' emotional bonding and closeness, whereas *adaptability* refers to their adaptability to change in response to external stressors. Based on this theoretical framework, partners who perceive greater family cohesion and who report greater adaptability (as reflected by lower family stress and individual psychological symptoms, and less problems with changing roles as function of illness) may be less likely to suffer from a spillover effect of the illness stress onto their relationship and may experience better relationship adjustment over time. Supporting this hypothesis, a recent review on parental distress related to childhood cancer revealed that early and intense

negative affectivity, as well as family stressors and family weaknesses at the time of diagnosis were among the best predictors of long-term individual distress in parents (Sultan et al., 2015). We aim to examine whether these results might be extended to marital adjustment. In particular, we predicted that less negative mood states (i.e., less anxiety, depression and fatigue) and better family cohesion (i.e., family cohesion and support) and adaptability (i.e., lower family stress, role ambiguity, role conflict, and role overload) at diagnosis would be associated with better marital adjustment at 2-years post diagnosis in parents of children diagnosed and treated for cancer. Gender differences in these associations were also examined, although no a priori hypotheses were put forward due to the lack of a previous empirical basis.

Methods

Participants and procedure

The current study is a secondary analysis of longitudinal data from the Québec cohort of children with acute lymphoblastic leukemia (QcALL cohort) and their parents (Marcoux et al., 2013; Marcoux, Robaey, Krajinovic, Moghrabi, & Laverdière, 2012). Children (18 years or less) with acute lymphoblastic leukemia (ALL) who were diagnosed (first occurrence) and treated at the Sainte-Justine University Health Centre, between February 1993 and September 1999, and their parents were consecutively recruited to participate in the study (138 families). T1 and T2 assessments were completed just after diagnosis (on average 2.5±2.5 weeks) and 2 years post diagnosis (104.3±2.7 weeks). (Marcoux et al., 2013; Marcoux et al., 2012). For the purpose of the present study, couples for whom only one of the two parents provided data were excluded (n = 25). Couples who were separated, divorced, or widowed at Times 1 or 2 were also

excluded (n = 29). The sample consisted of 84 intact couples at diagnosis, resulting in an overall participation rate of 61%. Parents individually completed self-report questionnaires at diagnosis (T1) and 2-years post diagnosis (T2), and were interviewed for demographic information. Only couples for whom data were available for both partners at T2 were included in our analyses, resulting in a final sample of 47 couples. Six couples did not participate at T2 as their child had deceased during the study period. Couples who participated at T1 only (n = 37) and those who participated at both time points did not significantly differ on any of the medical (child's diagnosis and treatment), sociodemographic (child's gender and age, parents' age and income) or study variables (mood and family well-being) at T1. All couples provided informed consent and the Institutional Review Ethics Board approved the research protocol. Demographic and medical information related to the illness are summarized in Table 1.

Measures

Marital adjustment

The Locke-Wallace Marital Adjustment Test (MAT) (Locke & Wallace, 1959) is a 15-item scale that has been widely used to measure marital adjustment (Reese, Somers, Keefe, Mosley-Williams, & Lumley, 2010; Woloski- Wruble, Dekeyzer Ganz, Jiang, Qiang, & Kadmon, 2012). It differentiates between individuals who are well adjusted in their relationship and those who are not. The MAT has excellent psychometric properties (α = .90) (Locke & Wallace, 1959) and has previously been used with couples of chronically ill children (Alderfer et al., 2008). Global scores range from 2 to 158, with higher scores reflecting better adjustment (Locke & Wallace, 1959). A clinical cut-off

score of 100 is used to identify significant marital distress (Cano, Gillis, Heinz, Geisser, & Foran, 2004; Reese et al., 2010).

Family functioning

The Family Well-Being Assessment (FWA) (Caldwell, 1988) assesses an individual's perception of family well-being on 12 dimensions (74 items), which are assessed on a continuum of well-being to stress. Items are rated on a 6-point Likert-type scale, with higher scores indicating higher stress on each dimension respectively. The FWA has acceptable psychometric properties, including an excellent reliability coefficient for the total scale (α = .90) (Caldwell, 1988) and it is particularly applicable and valid for families with chronically ill children (Caldwell, 1988). The choice of subscales included in the current study was based on the Circumplex Model of Marital and Family Systems. Family support and cohesion served as indicators of *Family cohesion,* whereas family stress, role ambiguity, role conflict, and role overload served as indicators of *Family adaptability. Family support* refers to the extent to which one feels that the family is there to take care of and support him/her (α = .86). *Family cohesion* refers to the extent to which one feels that one has both personal autonomy and a sense of belonging with the family (α = .67). *Family stress* refers to feelings of frustration and strain within the family (α = .80). *Role ambiguity* refers to family members' vague or unclear role expectations (α = .74). *Role conflict* refers to disputes regarding family members' roles and expectations (α = .68). *Role overload* refers to one's struggle in reaching role expectations with available resources (α = .79) (Caldwell, 1988). To facilitate interpretation, *Family support* and *Family cohesion* were reverse-coded for the

main analyses so as to reflect the actual label of the subscale (e.g., high scores on family support = higher family support).

Parental mood states

The Profile of Mood States-Bipolar Form (POMS-BI) (Lorr, McNair, & Heuchert, 2003) (72-item) uses adjectives or phrases to describe an individual's mood states in the past week (Lorr et al., 2003). Items are rated on a 4-point Likert-type scale (0 = *much unlike this*; 3 = *much like this*). Three subscales were selected as being most pertinent in the context of this study: (a) composed-anxious, (b) elated-depressed, and (c) energetic-tired. As per standard practice, raw scores were converted to T-scores (Lorr et al., 2003), and to facilitate interpretation, variables were reverse-coded so as to reflect greater endorsement of the negative aspects (e.g., anxious as opposed to composed). The POMS-BI has good to excellent reliability (α = .80 - .90) (O'Halloran, Murphy, & Webster, 2004), and test-retest reliabilities for the individual dimensions typically range from .33 to .72 (Lorr et al., 2003).

Statistical analyses

Preliminary analyses

The distributions of all variables were examined for normality. Non-linear transformations were successfully applied to skewed variables (skew values > 1): marital adjustment was negatively skewed and subjected to a square root transformation, and tiredness was positively skewed and subjected to a logarithmic transformation. All other variables were normally distributed. There was no missing data. A series of paired t-tests and repeated measures ANOVAs and MANOVAs, as well as bivariate correlations were used to initially test the associations between mothers' and fathers' variables, and identify

potential control variables among the medical and sociodemographic variables. Means and standard deviations for the study variables are presented in Table 2.

Main analyses

To assess dyadic associations among family functioning, mood, and marital adjustment in parents, we used the Actor-Partner Interdependence Model (APIM) as our core data analysis strategy (with SPSS MIXED MODELS). The APIM is a modified regression-based technique, which allows for prediction of outcome variables among dyads (see Figure 1) (Kenny, Kashy , & Cook, 2006). This multilevel modeling approach has inherent advantages over traditional regression analyses (Hall, 2010b): a) accounting for the non-independence of couple data, b) simultaneously testing both actor effects and partner effects, and c) testing gender differences in the strength of actor and partner effects (Kenny et al., 2006). An *actor effect* refers to the effect of an individual's own family functioning and mood on their own marital adjustment. A *partner effect* refers to the effect of an individual's own family functioning and mood on their partner's marital adjustment.

Multilevel linear models were conducted to predict partners' marital adjustment at 2-years post diagnosis from partners' family functioning and mood scores at diagnosis ($p < .05$). Separate models were conducted for each predictor (family functioning and mood variables) at the time of diagnosis. As recommended by Kenny et al. (Kenny et al., 2006), all predictor and outcome variables were standardized prior to conducting these analyses. Partners' predictors (family functioning or mood states) and the error terms for partner's marital adjustment were allowed to correlate to account for the non-independence of partners' scores (see Figure 1 for an example of such a model). To explore potential

gender differences, gender and the interaction between gender and predictors were included in all analyses. A significant interaction term indicates a significant gender difference in the strength of an actor or partner effect. All analyses also controlled for mothers' and fathers' marital adjustment at diagnosis.

Results

Preliminary analyses

Preliminary descriptive analyses revealed that 25.5% of mothers and 21.3% of fathers reported significant marital distress at the time of diagnosis, as indicated by a score below 100 on the MAT. Two years later, 36.2% of mothers and 42.6% of fathers met this cut-off. Mothers' and fathers' marital adjustment scores at diagnosis (T1) were positively correlated with their own marital adjustment scores 2-years post diagnosis (T2) (Table S1). Mothers' marital adjustment at diagnosis was also positively associated with fathers' marital adjustment both at diagnosis ($r = .53$ $p < .001$) and 2-years post diagnosis ($r = .55$, $p = .022$).

Preliminary analyses revealed no significant associations between personal or clinical variables (patient's gender and age, initial diagnosis, number of hospitalisation days, use of radiotherapy, treatment protocol, relapse during the study) or sociodemographic variables (parents' age, personal income, years of education) and marital adjustment. Therefore, none of these variables were controlled in the main analyses. Paired *t*-tests showed that mothers perceived less family cohesion ($t(46) = 3.10$, $p = .003$) than fathers, whereas fathers reported being more tired ($t(46) = 2.95$, $p = .005$) and perceived more role conflict than mothers ($t(46) = 2.05$, $p = .046$) at the time of

diagnosis. No other gender difference was found on the family well-being, mood or marital adjustment variables.

Main analyses

Standardised regression coefficients from APIM models for all significant actor, partner, and gender effects are presented in Table 3. All models controlled for T1 marital adjustment. Several actor effects were found. For mothers, increased marital adjustment at T2 was associated with her perception of greater family support, less role conflict, and less role overload at the time of diagnosis. For fathers, increased marital adjustment at T2 was associated with his perception of less role conflict, greater role ambiguity, and being more tired at diagnosis. Significant gender differences were found for all of the aforementioned actor effects, except for role overload, which did not significantly differ between mothers and fathers. As role conflict was a significant predictor in both mothers and fathers, the gender difference indicates that this effect was stronger for mothers. One partner effect was found. For fathers, better marital adjustment at T2 was associated with mothers perceiving less role conflict at diagnosis. There was a significant gender difference for this partner effect (Figure 1).

Discussion

In this dyadic longitudinal study, we examined evolutions of marital adjustment over two years post diagnosis in both parents of children treated for ALL, in relation to their mood and family functioning at the time of diagnosis. An important finding was that positive changes in marital adjustment over time were associated with different characteristics of family functioning and mood for mothers and fathers. We also found

that responses of mothers just after diagnosis were also associated with changes in fathers' marital functioning.

In our sample, mothers and fathers reported similar levels of marital adjustment on both occasions. This is coherent with findings from a meta-analysis on parental distress, and family and marital functioning in parents of children with cancer (Pai et al., 2007). A substantial portion of parents reported clinically significant marital distress at diagnosis (25.5% of mothers and 21.3% of fathers) and this increased over time (36.2% of mothers and 42.6% of fathers). These percentages are congruent with findings from other studies of parents in the pediatric cancer context (Dahlquist & Czyzewski, 1993; Dahlquist et al., 1996), and they highlight that marital distress is common in this population, even two years after the initial diagnosis. These findings also underscore the importance of conducting research aimed at better understanding the determinants of relationship adjustment in parents in order to be able to respond to their specific needs in the initial and later stages of the illness.

Significant differences between fathers and mothers were found on family functioning and mood. Overall, these differences were consistent with previous comparisons showing that fathers experience acute gender role conflict during the treatment phase reflected by higher role conflict (Hall, 2010a; Sultan et al., 2015). In fact, fathers have reported that the role of economic supporter in the family is often at odds with the emotional support needed by family members (McNeill, 2007). This role conflict may also translate into elevated tired mood levels, as was reflected by the higher levels found in fathers than in mothers.

When exploring longitudinal associations, mothers' increased marital adjustment over time was associated with greater reported family support, less perceived role conflict, and less role overload at the time of diagnosis. This indicates that mothers perceiving that they were supported by their family at diagnosis (i.e., family cohesion) appeared to serve as protective factors for their future marital adjustment, which was not the case in fathers. Conversely, the experience of role conflict after diagnosis, such as perceiving more conflict regarding family roles and their ability to meet these role expectations (i.e., low family adaptability), is related to a deterioration in mothers' marital adjustment over time. Mothers' perception of role conflict and role overload within a family with an ill child may be an additional source of stress which aggravates their initial distress, and could later spill over into their relationship (Hall, 2010b). We found this to be the case to a larger extent in mothers than in fathers. These findings are coherent with the Circumplex Model of Marital and Family Systems as both aspects of cohesion and adaptability appear to play a role in future adjustment (Olson et al., 1983). Mothers' marital adjustment was not predicted by their partners' variables, which could indicate that their partner's perceived family functioning may not be so relevant for their future marital adjustment. It is possible that their perceived support from the family unit might be sufficient in ensuring that they are well adjusted in their relationship with their partner. This makes sense given that mothers of children with cancer usually seek more social support than fathers (Goldbeck, 2001). It could also be that patterns of support offered by fathers in the context of their perceived role conflict are not optimal and thus do not translate into mothers' future adjustment (Hall, 2010a).

In fathers, associations observed with role conflict are consistent with those observed in mothers. Yet, positive changes in marital adjustment were associated with more role ambiguity. At first sight this may appear counter-intuitive. However, it is very coherent with the fact that fathers may perceive greater role changes following diagnosis. While mothers are typically the primary caregivers to the ill child, fathers are often required to take care of the other children at home and maintain daily routines, which may be unusual tasks for them (Goldbeck, 2001), and mothers are more often at the forefront of the interactions with the hospital. Previous data has suggested that fathers of children with cancer experience considerable gender-role adaptation (Hall, 2010a). Our findings suggest that confronting family roles and reorganizing said roles in the early stages of the illness could strengthen their relationship with their partner over time. Surprisingly, we found that those fathers showing higher fatigue at diagnosis also experienced less marital distress over time. Fathers' fatigue could reflect higher active involvement and supportive behaviours, which would positively impact their perceived relationship later. As reflected in the correlations in Table S1, tiredness was not associated with anxiety or depressed mood and is thus probably not a marker of negative affect here.

Interestingly, beyond their own perception of family functioning, fathers' marital adjustment over time was also predicted by their partners' perception of family functioning. When mothers experienced heightened role conflict around the time of diagnosis, fathers also reported deteriorations in marital adjustment two years later (i.e., partner effect). It is possible that mothers would express dissatisfaction and frustrations, or make demands to have their needs met regarding each parent's role in the family. Such

demands may create more relationship conflict and explain fathers' deteriorations in relationship adjustment later. Indeed, the literature on spousal involvement in chronic illness indicates that the unsupportive behaviours exhibited by the spouse appear to be particularly influential in understanding adjustment (Manne, Ostroff, Winkel, Grana, & Fox, 2005).

Our findings have several implications for clinical research and practice in pediatric oncology. During the treatment of leukemia, it appears that both parents may benefit from interventions targeting role-related burden and stress, for instance by insisting on solving problems raised by multiple role issues, as well as maintaining one's vitality and protecting one's long-term resources. Recent intervention models have addressed such practical problem-solving skills in parents (Sahler et al., 2005). Interventions stressing family support with mothers also appear to be highly relevant. Intervention models have addressed communication issues and dyadic adjustment in parental couples confronting childhood cancer. Greater awareness that partners might react differently following their child's illness could also potentially lead to greater empathic understanding and support within couples. (Kazak et al., 2005; Li & Loke, 2014).

We must acknowledge the limitations of the current study. First, our findings reflect the experience of a limited sample of primarily well-adjusted couples from a single patient-centered childhood cancer center within the public health system. They may not be representative of highly distressed couples or those with greater financial burden. The high homogeneity of the clinical condition within this sample (pediatric ALL) also limits the transferability of our conclusions to other cancer types and stages.

The small sample size may also have reduced statistical power and our ability to detect significant associations among variables. Lastly, although we controlled for baseline marital adjustment in predicting long-term adjustment, the observational nature of this study precludes making any definite conclusions about causation.

To conclude, in a sample of 47 parental couples confronted with childhood leukemia and using the APIM analysis framework, we found independent actor effects in mothers and fathers showing that improvements in perceived marital adjustment were differentially predicted by their perceived family functioning and mood at time of diagnosis. Our findings also support the idea that marital adjustment of fathers over time would partly depend on mothers' initial perceived family functioning. Future studies should further investigate how parents' roles are experienced when they are confronted with childhood cancer as these aspects may influence their marital well-being in the long term.

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Table 1. *Sample description (n = 47 couples).*

Child's characteristics	**M (SD) or N (%)**	
Age at diagnosis, years	5.92 (4.10)	
Length of initial hospitalization (days)	30.51 (8.58)	
Sex of child		
Boys	31 (66.0)	
Girls	16 (34.0)	
ALL risk status		
Standard risk	20 (42.6)	
High risk	27 (57.4)	
Treatment protocol		
DFCI 91-01	19 (40.4)	
DFCI 95-01	28 (59.6)	
Cranial radiation therapy		
No	10 (21.3)	
Yes	37 (78.7)	
Relapse during the study		
No	43 (91.5)	
Yes	4 (8.5)	
Parents' characteristics	**Mothers M (SD) or N (%)**	**Fathers M (SD) or N (%)**
Age at diagnosis, years	34.23 (4.64)	36.95 (5.46)
Years of education	13 (2.22)	13 (2.93)
Financial income, at diagnosis		
Not applicable	3 (7.3)	0 (0.0)
< 29,999	27 (65.9)	16 (39.0)
30,000 - 49,999	11 (26.8)	17 (41.5)
≥ 50,000	0 (0.0)	8 (19.5)
Number of children living at home	1.83 (2.22)	

Table 2. *Mean (standard deviations) for marital adjustment, family functioning, and mood variables for mothers and fathers*.

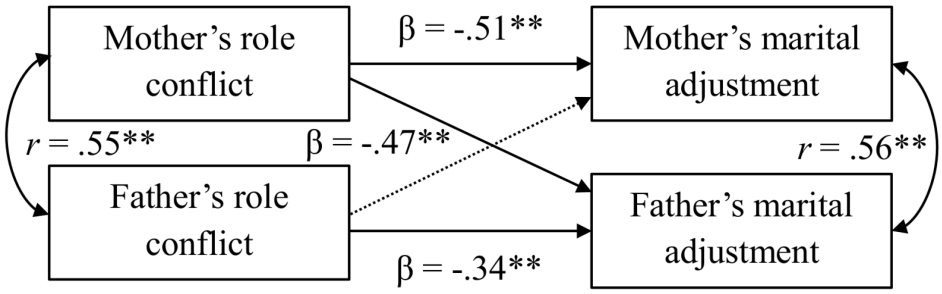
Variable	Mothers	Fathers
Marital adjustment		
Marital adjustment (T1)	114.21 (21.38)	112.70 (26.02)
Marital adjustment (T2)	104.38 (30.34)	103.28 (27.78)
Family functioning (T1)		
Family stress	2.83 (1.14)	2.91 (1.10)
Family support	2.29 (1.00)	2.38 (.81)
Family cohesion	**2.15 (.81)**	**2.49 (.71)**
Role conflict	**2.70 (1.02)**	**2.98 (.96)**
Role overload	2.77 (1.29)	2.58 (.93)
Role ambiguity	1.89 (.75)	2.08 (.75)
Mood (T1)		
Anxious mood	50.26 (9.81)	48.89 (9.59)
Depressed mood	53.02 (10.46)	51.98 (11.77)
Tired mood	**50.47 (10.56)**	**45.36 (6.05)**

Note. Higher scores on the marital adjustment variable indicate better adjustment. Family functioning variables are scored on a continuum of well-being to stress, with higher scores indicating higher stress on each dimensions respectively. Lower scores on the mood variables indicate greater endorsement of the negative mood (anxious, depressed, and tired, respectively). Bolded means indicate significant gender differences.

Table 3. *Summary of actor, partner and gender effects as identified by APIM models predicting marital adjustment at 2-years post diagnosis with family functioning and parental mood states at diagnosis*.

	Actor effect			Partner effect		
Predictors (at diagnosis)	**Father**	**Mother**	**Gender difference**	**Father**	**Mother**	**Gender difference**
Family support		β = .487	p = .021			
Role conflict	β = -.341	β= -.505	p < .001	β = -.474		p = .022
Role overload		β= -.366	p = .131			
Role ambiguity	β = .445		p < .001			
Tired	β = .383		p = .009			

Note. Family support, Family cohesion, Role conflict, Role overload, and Role ambiguity were measured with the FWA. Tired was measured with the POMS-BI. To facilitate interpretation here, Family support and Tired mood were reverse-coded so as to reflect the actual label of the subscale (e.g., high scores on family support = higher family support). All analyses controlled for mothers' and fathers' marital adjustment at diagnosis. For clarity, only significant regression coefficients are displayed.



Note. ** p < .01, * p < .05; the dashed line represents a non-significant association.

Figure 1. *Actor-partner interdependence model (APIM) predicting marital adjustment at 2-years post diagnosis from role conflict at diagnosis (n = 47 couples.)*

Table S1. *Correlations between variables for mothers and fathers at diagnosis (T1) and 2-years post diagnosis (T2*).

	1	2	3	4	5	6	7	8	9	10	11
1. Marital adjustment (T1)	.53**	.51**	-.24	.54**	.56**	-.36*	-.24	-.31*	-.08	.13	-.27
2. Marital adjustment (T2)	.48**	.55**	-.28*	.62**	.47**	-.32**	-.52**	-.38**	-.006	.16	-.26
3. Family stress (T1)	-.10	-.01	.53**	-.41**	-.43**	.47**	.33*	.05	.44**	-.23	-.15
4. Family support (T1)	-.32*	.15	-.23	.53**	.63**	-.74**	-.53**	-.51**	-.12	.27	-.27
5. Family cohesion (T1)	.48**	.10	-.48**	.42**	.59**	.64**	-.53**	-.50**	.02	.03	-.27
6. Role conflict (T1)	-.36*	-.05	.42**	-.38**	-.58**	.56**	.58**	.52**	-.09	.21	.13
7. Role overload (T1)	-.36*	-.24	.39**	-.29*	-.61**	.62**	.52**	.60**	-.19	-.06	.11
8. Role ambiguity (T1)	-.36*	.24	.13	-.45**	-.41**	.54**	.33*	.14	.16	.07	.20
9. Anxious mood (T1)	.04	-.02	-.04	-.11	-.04	-.14	.00	-.10	-.04	.43**	.14
10. Depressed mood (T1)	-.27	-.05	.07	-.30*	-.26	.20	.15	.29*	.26	.03	.08
11. Tired mood (T1)	.06	.34*	.01	.10	.15	-.03	-.03	.03	-.05	-.11	.09

Note. Mothers' bivariate correlations are above the diagonal and fathers' bivariate correlations are below the diagonal. Interrelationships between the partners' variables are displayed on the diagonal. ** $p < .01$, * $p < .05$

Article 2

Cancer-related effects on relationships, long-term psychological status and relationship satisfaction in couples whose child was treated for leukemia: A PETALE study

Burns, W., Péloquin, K., Rondeau, É., Drouin, S., Bertout, L., Lacoste-Julien, A., Krajinovic, M., Laverdière, C., Sinnett, D., & Sultan, S. (under review). Cancer-related effects on relationships, long-term psychological status and relationship satisfaction in couples whose child was treated for leukemia: A PETALE study. *Submitted to PLoS ONE*.

Running head: CANCER-RELATED EFFECTS ON RELATIONSHIPS AND LONG-TERM ADJUSTMENT IN COUPLES

TITLE:

Cancer-related effects on relationships, long-term psychological status and relationship satisfaction in couples whose child was treated for leukemia: A PETALE study

AUTHORS:

Willow Burns, MA (PhD Candidate), Department of Psychology, University of Montreal, Montreal, Quebec, Canada; Research Center, Sainte-Justine University Health Center, Montreal, Quebec, Canada.
Katherine Péloquin, PhD, Department of Psychology, University of Montreal, Montreal, Quebec, Canada.
Émélie Rondeau, MA, Research Center, Sainte-Justine University Health Center, Montreal, Quebec, Canada.
Simon Drouin, PhD, Research Center, Sainte-Justine University Health Center, Montreal, Quebec, Canada.
Laurence Bertout, Research Center, Sainte-Justine University Health Center, Montreal, Quebec, Canada.
Ariane Lacoste-Julien, Research Center, Sainte-Justine University Health Center, Montreal, Quebec, Canada.
Maja Krajinovic, MD, Department of Pediatrics, University of Montreal, Montreal, Quebec, Canada; Research Center, Sainte-Justine University Health Center, Montreal, Quebec, Canada.
Caroline Laverdière, MD, Department of Pediatrics, University of Montreal, Montreal, Quebec, Canada; Research Center, Sainte-Justine University Health Center, Montreal, Quebec, Canada.
Daniel Sinnett, PhD, Department of Pediatrics, University of Montreal, Montreal, Quebec, Canada; Research Center, Sainte-Justine University Health Center, Montreal, Quebec, Canada.
Serge Sultan, PhD, Department of Psychology & Pediatrics, University of Montreal, Montreal, Quebec, Canada; Research Center, Sainte-Justine University Health Center, Montreal, Quebec, Canada.

CONTACT INFORMATION OF CORRESPONDING AUTHOR:

Serge Sultan, PhD
Research Center, Sainte-Justine University Health Center
3175, Chemin de la Côte-Sainte-Catherine
Montreal, Quebec, Canada, H3T 1C5
Tel. + 1 (514) 343-6111 (20727)
Email : serge.sultan@umontreal.ca

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Abstract

Objectives: Follow-up studies suggest that the psychosocial impact of pediatric cancer on parents often extends beyond the end of their child's cancer treatments, and parents can continue to experience both individual and relationship effects. In a long-term study of parents of children who were treated for acute lymphoblastic leukemia (ALL), we aimed to: 1) describe parents' adjustment (psychological distress, relationship satisfaction; 2) describe the perceived impact of cancer on couples' relationship, and; 3) identify to what extent the perceived impact of cancer on the couple is related to both parents' long-term adjustment.

Methods: Parents of childhood ALL survivors (n = 103 couples) were surveyed as part of a cohort recall (PETALE cohort). Both parents completed questionnaires exploring adjustment (Brief Symptom Inventory-18, Dyadic Adjustment Scale) and perceived impact of cancer on the relationship (Impact of Cancer on the Couple). Mothers' and fathers' scores were compared using MANOVAs. We also examined the degree to which a parent's perceived changes in relationship dynamics following their child's cancer were associated with their own current adjustment (actor effects), and their partner's current adjustment (partner effects) using Actor-Partner Interdependence Model (APIM).

Results: Frequencies of current distress were normative in parents (mothers/fathers): general distress (6.8/7.8%), anxiety (5.8/6.8%), depression (2.9/6.8%), somatization (13.6/9.7%), and relationship distress (21.4/20.4%). Mothers and fathers typically agreed on their reported relationship satisfaction, and the perceived nature of relationship changes following the illness. Dyadic analyses indicated that whereas mothers'

adjustment was related to their own perceived relationship changes, fathers' adjustment was primarily related to their partner's perceptions.

Conclusion: In long-term stable couples, mothers may act as an influential bridge connecting the illness experiences of survivors and fathers. This could explain why mothers' perceptions of relationship changes were related to their partners' long-term adjustment, which was not the case for fathers.

Introduction

Childhood cancer has been identified as a long-term vulnerability factor for parents' well-being at both the individual level (Hardy et al., 2008; Kinahan et al., 2008; Klassen et al., 2007; Ljungman et al., 2014; Malpert et al., 2015) and the level of parents as a couple (Burns et al., 2016; da Silva, Jacob, & Nascimento, 2010; Van Schoors, Caes, Alderfer, Goubert, & Verhofstadt, 2016; Wiener et al., 2016). Although reports have documented individual distress levels in parents, few have compared both parents in the couple and explored dyadic interrelations within couples. In addition, no studies have systematically surveyed the perceived impact of cancer on parents' relationship and how this may explain the current adjustment of parents several years after the illness has subsided. In this study, we aimed to assess long-term psychological status and relationship satisfaction in parents of childhood leukemia survivors, explore their perceived impact of cancer on their relationship, and how this impact may explain both parents' current individual and relationship adjustment. A recent review on parents of childhood cancer survivors suggested that although most parents reported normal ranges of psychological distress, a significant subgroup reported clinically significant distress (Ljungman et al., 2014), with 21-44% of parents reporting severe posttraumatic stress symptoms (PTSS). In contrast, within a recent cross-sectional study of parents of long-term acute lymphoblastic leukemia (ALL) survivors, clinically significant anxiety, depression, and posttraumatic stress were reported by 7.1%, 3.1%, and 3.9% of parents, respectively (Malpert et al., 2015). Thus, there appears to be great variability in the proportion of parents reporting significant distress as indicated by different studies. Consequently, this might suggest that select factors may explain these varying rates of

distress. Factors associated with heightened distress in parents have been identified. First, the time elapsed since the child's diagnosis has been reported as a factor associated with parental distress, such that distress typically decreases as more time passes (Ljungman et al., 2014; Sultan, Leclair, Rondeau, Burns, & Abate, 2016). Second, parents' use of maladaptive coping strategies earlier in the illness and their child's poor adjustment, have also been found to predict parents' long-term or late effects (Ljungman et al., 2014). Third, a constructive social context surrounding the illness, such as better family functioning and availability of social support, can also help attenuate parents' distress (Ljungman et al., 2014; Sultan et al., 2016). Indeed, parents in conflictive families tend to report more anxiety, depression, and PTSS, while cohesive families tend to report less depression symptoms (Ozono et al., 2010). Additionally, parents' gender was a significant factor, with mothers reporting more distress especially early in the illness trajectory (Ljungman et al., 2014; Sultan et al., 2016).

Another factor, which may explain the current psychological status of parents, is the impact of cancer on their relationship. A few select reports have investigated the impact of cancer on the relationship of the parental couple. Some couples emphasize that the illness had a positive impact on their relationship (e.g., greater trust, communication, support, and emotional closeness), whereas others emphasize its negative impact (e.g., deteriorations in sexuality; da Silva et al., 2010; Lavee & Mey-Dan, 2003; Van Schoors et al., 2016; Wiener et al., 2016). To date only two empirical studies have directly assessed which aspects of the parents' relationship changed over the illness trajectory and the extent to which the cancer experience challenged or tested their relationship (Lavee & Mey-Dan, 2003; Wiener et al., 2016). However, these studies found contrasting results.

One multicenter cross-sectional survey of parents of children currently being treated for cancer (who were at least 3 months post-diagnosis) or in follow-up care (completed cancer treatment within past 3 years; $N = 192$ parents; 122 mothers and 70 fathers) found that a third of parents experienced deterioration in their relationship quality and low dyadic adjustment, with more than half claiming that their relationship as a couple had been challenged following the illness (Wiener et al., 2016). A smaller cross-sectional study ($N = 35$ couples) found that spouses whose child with cancer was between 1 to 7 years post-diagnosis experienced strengthened communication, deteriorations in sexuality, and no change on relationship dimensions of conflict resolution, leisure activities, and division of household labour (Lavee & Mey-Dan, 2003). A recent review on couples' functioning following their child's cancer diagnosis parallels these mixed findings on conflict reported by both qualitative and quantitative studies (Van Schoors et al., 2016). Notably, no reports have explored the impact of cancer on important relationship areas such as partner support and intimacy. As exemplified in these reports, parents' adjustment to cancer has most often been studied with each partner analyzed separately. For instance, a longitudinal study of parents of children with cancer found that for both mothers and fathers their marital adjustment at follow-up was partially explained by their spouse's marital satisfaction scores. Although this study can boast that it considered the association between one spouse's marital satisfaction and the other spouse's marital adjustment, this study was not truly dyadic in its design. Hierarchical multiple regression analyses were conducted separately for mothers and fathers. Interdependence in the couples' data and potential gender differences in these associations were also not statistically accounted for or tested (Dahlquist, Czyzewski, &

Jones, 1996). Nevertheless, this study was conducted at 2 and 20 months post-diagnosis; hence it does provide a basis for assuming that relationship dynamics during the child's treatment could also be associated with parents' relationship adjustment in the survivorship period.

Recent studies have started to address dyadic interrelationships in parents of children with cancer. However, only four empirical studies in the field of pediatric cancer have been conducted thus far (Burns et al., 2016; Compas et al., 2015; Hall, 2010, 2010b), and of these only one had a longitudinal design (Burns et al., 2016). Notably, that study found that mothers' adjustment 2 years after their child's ALL diagnosis was associated with their own perception of family support, role conflict, and role overload at diagnosis. Fathers' adjustment was associated with both their own perceptions (role conflict, role ambiguity, being tired) and their partner's perception (role conflict) at diagnosis (Burns et al., 2016). To date, no such strategy has been used to articulate the perceived relationship impact of cancer on partners' current emotional adjustment or relationship quality.

Our research objectives were threefold. We aimed to: 1) Describe psychological and relationship adjustment (psychological distress, relationship satisfaction) of mothers and fathers whose child was treated for acute lymphoblastic leukemia. To complement previous reports, we wished to study parents' adjustment long after children's remission (>5 years post-diagnosis). 2) Describe the perceived impact of cancer on these couples using a systematic approach of couples' functioning including core relationship dimensions such as intimacy, partner support, sexuality, conflict, or shared time and activities. 3) Identify to what extent the perceived impact of cancer on the couple is

related to both parents' long-term adjustment (psychological distress, relationship satisfaction). To bridge the gap in the existent literature, we examined actor (i.e., the effect of one's perceived impact of cancer on their own adjustment) and partner (i.e., the effect of a parent's perceived impact of cancer on the other parent's adjustment) effects, as well as gender differences in these effects.

Materials and methods

Participants

Participants were parents of childhood ALL survivors from the PETALE cohort (Marcoux et al., 2017). Childhood ALL survivors that were diagnosed and treated at Sainte-Justine University Health Centre (SJUHC, Montreal, Canada), with DFCI protocols 87-01 to 2005-01, and their parents were recruited to participate in this long-term follow-up study (224 families). In order to be eligible to participate in the PETALE study, ALL survivors had to: (a) be less than 19 years of age at the time of their diagnosis; (b) not having received a transplant and not having experienced relapse or a second cancer, and; (c) be at least 5-years post-diagnosis at the time of recall.

Given the objectives of this study, families were excluded if the data was only available for the survivor but not their parents (n = 31) or for only one parent (n = 84). Parents who were not caring for their child during the illness or couples that were separated either during their child's cancer treatments or at the time of cohort recall were also excluded (n = 6). The final sample was thus comprised of 103 'intact couples' (i.e., stable couples that were together both during their child illness and at the time of this cohort recall; see **S1 Fig**), resulting in a final participation rate of 46% (103 / 224 families).

On average, mothers and fathers were 51 and 54 years old respectively and survivors were 22 years old at the time of recall. Survivors were also on average 15 years post-diagnosis and slightly more than half reported known late-adverse effects at the time of assessment (**Table 1**).

Procedures

The research coordinator or clinical research assistant invited eligible families from the PETALE cohort to participate in this recall study by phone. The ALL survivors who agreed to participate came to the hospital for a day of testing. Parents completed a series of questionnaire on site if they accompanied their child to the hospital, or at home and returned them by mail if they were not present. Both parents were invited to participate and were asked to complete their questionnaires independently. The research coordinator called the parents for a follow-up if the questionnaires were not returned within 3 weeks. Data were checked for clinically significant distress and appropriate referrals were made when deemed necessary in order to comply with ethical standards. The research coordinator or clinical research assistant would call parents to collect missing data. Survivors' medical information was collected from patients' medical records. The research coordinator collected survivors' socio-demographic information during the day of testing. All participants provided informed consent and the Institutional Review Ethics Board at SJUHC approved the study. Further description of this cohort is available in a previous report (Marcoux et al., 2017).

Measures

Brief Symptom Inventory-18 (BSI-18; Derogatis, 2000).

The Brief Symptom Inventory is an 18-item self-report questionnaire, assessing psychological distress (Derogatis, 2000). Previous studies have also specifically used this measure with cohorts of adolescent and adult survivors of childhood cancer (Gianinazzi et al., 2013; Recklitis et al., 2006), as well as with their parents (Leclair, Carret, Samson, & Sultan, 2016). It includes three symptom dimensions: Anxiety (α = .86*)*, Depression (α = .88*)*, and Somatization (α = .74), as well as a total score, the Global Severity Index (GSI), which reflects an individual's global level of distress (α = .93). Respondents are asked to report on their symptoms in the past 7 days. They can be classified with their standardized T-scores as either being at a high risk for psychological distress symptoms (i.e., *positive caseness*; $T_{GSI} \geq T_{63}$ or $T_{2DIMENSIONS} \geq T_{63}$) or not being at any apparent risk (i.e., *negatives caseness*; Derogatis, 2000).

Dyadic Adjustment Scale (DAS-4) (Sabourin, Valois, & Lussier, 2005).

The abbreviated Dyadic Adjustment Scale (DAS-4; Sabourin et al., 2005) evaluates current relationship satisfaction using four items (α = .84). Although this exact version of the Dyadic Adjustment Scale (DAS-4) has been widely cited in the field of couples' research (Arden- Close, Moss- Morris, Dennison, Bayne, & Gidron, 2010; Brassard, Lussier, & Shaver, 2009; Péloquin & Lafontaine, 2010), it has not previously been used with parents of children with cancer or parents of childhood cancer survivors. However, studies on couples that have a partner with cancer have used the original 32-item Dyadic Adjustment Scale (DAS; Aerts et al., 2015; Cochrane, Lewis, & Griffith, 2011; Hamilton, Van Dam, & Wassersug, 2016) or brief versions (Götze et al., 2017;

Green, Wells, & Laakso, 2011; McLean, Walton, Matthew, & Jones, 2011; Walker, King, Kwasny, & Robinson, 2016). To date, only one empirical study has used a form of the Dyadic Adjustment Scale (14-items) with parents of children with cancer that are either actively being treated or have completed cancer treatments (Wiener et al., 2016). A global DAS-4 score is calculated by summing the four items (range 0-21), with a higher score thereby suggesting greater relationship satisfaction. The DAS-4 has been found to effectively classify couples as clinically distressed (DAS < 13) or non-distressed (DAS ≥ 13), as well as predict couples' dissolution over time (Sabourin et al., 2005).

Impact of Cancer on the Couple (Péloquin & Sultan, 2013).

The Impact of Cancer on the Couple is a brief survey composed of 7-items that was developed specifically in the context of the current study to assess the perception of changes in several relationship dimensions following a child's cancer diagnosis (Péloquin & Sultan, 2013). The first few items of the questionnaire were simply used for contextualization and screening purposes in this study (e.g., screening out parents who were separated, divorced, widowed or in a relationship with another partner who was not the parent of the childhood ALL survivor). Parents were asked to reflect back on the time that their child was in treatment and to rate the impact of their child's illness on six dimensions of their relationship with their partner: Intimacy, Quality of partner support, Sexuality, Conflict, Time spent together and activities, and Relationship satisfaction. Each relationship dimension is rated on a continuum ranging from "*1 = very negative effect*" to "*7 = very positive effect*". A score of 4 reflects "*no change"*. When parents report a negative effect (scores 1 to 3), they are asked to indicate the extent to which these negative effects persisted once cancer treatment ended ("*The negative effects*

disappeared immediately"; "The negative effects remained but faded over time"; "The negative effects were permanent"). To describe the nature of the changes experienced by parents, dimension scores were also classified into three main categories: negative change (scores of 1 to 3), no change (score of 4), and positive change (scores of 5 to 7). Parents were also asked to assess the Overall perceived impact of their child's illness on their relationship, and quantify this change on a 1 to 7 scale, ranging from *"1 = this period has distanced us/has been detrimental to our relationship"* to *"7 = this period brought us closer/strengthened our relationship."* A score of 4 on this particular item signifies *"this period had no effect on our relationship."* For the purposes of consistency and ease of visual representation, the same classifications as above were used to denote negative change, no change and positive change on this item. The scale showed good reliability (α = .84). The original French questionnaire (**S1 File**) and a translated English version (**S2 File**) are available for download as supplementary files to this article.

Statistical analyses

The distributions of all variables were assessed for normality. For Objectives 1 and 2 no transformations were applied. For Objective 3, non-normally distributed variables (skewness and kurtosis > 1) were subjected to the following non-linear transformations: a reflection and square root transformation on Quality of partner support (Impact of Illness on the Couple), a square root transformation on Global distress (Global Symptom Index - BSI-18), and inverse transformations on Anxiety, Depression, and Somatization (BSI-18). Depression variables were severely skewed, and parents in three couples had extreme depression scores (Z score > 3.5). Data from these three couples were retained in all analyses for Objectives 1 and 2, because the emphasis was primarily

descriptive. However, due to normality concerns they were excluded from the dyadic analyses in Objective 3. In the remaining couples, even after inverse transformations on both partners' depression variables were applied the resulting distributions were still slightly skewed but since these distributions more closely approached normality these inverse transformations were used in the dyadic analyses. The remaining variables were all normally distributed. There was no missing data. To detect possible control variables among clinical and demographic variables for Objective 3 (age of child at diagnosis, age of parents at diagnosis, age of survivor at follow-up, age of parents at follow-up, relationship length, time elapsed since diagnosis, time elapsed since end of treatment, sex of child, ALL risk group, use of radiotherapy, long-term complications), we conducted bivariate correlations and repeated-measures MANOVAs (where gender served as a repeated-measure for the couple). Given that no significant associations were found no covariates were included in the main analyses.

Objective 1: Description of parents' long-term adjustment

In order to compare mothers and fathers on adjustment variables, a repeated-measures MANOVA was conducted, where gender served as a repeated-measure for the couple. To assess the degree to which mothers and fathers resemble each other on adjustment variables, intra-class correlation coefficients (ICCs) were also calculated. ICC values were categorized as either: poor (ICC < .40), fair (ICC = .40-.59), good (ICC = .60-.74), or excellent (ICC = .75–1.00; Cicchetti, 1994). Next, we calculated the proportion of mothers and fathers reporting clinically significant scores on each adjustment variables. McNemar tests were used to compare the proportion of mothers and fathers meeting criteria for positive caseness.

Objective 2: Description of perceived impact of cancer on the couple

The same strategy was applied to compare mothers' and fathers' perceived changes in their relationship. Wilcoxon tests and bar chart comparisons (see **Fig 1**) were used to compare the proportion of mothers and fathers that reported each type of relationship dimension change (negative change, no change, positive change).

Objective 3: Dyadic models for long-term adjustment

Dyadic associations among cancer-related relationship changes, psychological distress, and relationship satisfaction were examined using the Actor-Partner Interdependence Model (APIM) with SPSS MIXED MODELS, a modified regression-based technique which allows for prediction of outcome variables among dyads (see **Fig 2**; Kenny et al., 2006). This multilevel modeling approach has several advantages over traditional regression analyses: a) accounting for the non-independence of couples' data; b) simultaneously testing both actor and partner effects, and; c) testing gender differences in the strength of actor and partner effects (Kenny et al., 2006). Analyses were conducted to predict parents' current relationship satisfaction and psychological distress (Global Severity Index, Anxiety, Depression, and Somatization symptoms) from both partners' perception of relationship changes following their child's cancer treatments (Impact of Cancer on the Couple). We conducted separate models for each predictor (dimensions of relationship changes), and each outcome variable (see **Fig 2** for an example of one such APIM model). To explore potential gender differences, gender and the interaction between gender and predictors were included in all analyses. A significant interaction term indicates a significant gender difference in the strength of an actor or partner effect. Significance levels of $p < .05$ were set for all dyadic analyses.

Results

Objective 1: Description of parents' long-term adjustment

Results of the overall MANOVA showed that mothers and fathers significantly differed on their level of relationship satisfaction and psychological distress ($F(5, 98) = 5.70$, $p < .001$, $n_p^2 = .23$), with fathers reporting greater depression symptoms than mothers. No gender differences were found for relationship satisfaction, global distress, and symptoms of anxiety and somatization (**Table 2**). Poor agreement between parents was found on global distress and all psychological distress symptoms (i.e., anxiety, depression, and somatization). In contrast, mothers and fathers exhibited excellent agreement with respect to their reports of relationship satisfaction. We found that 21.4% of mothers and 20.4% of fathers reported clinically significant relationship distress (non-significant difference; **S1 Table**). A minority of parents scored within the clinical range on global distress (6.8% of mothers and 7.8% of fathers), anxiety (5.8% of mothers and 6.8% of fathers), depression (2.9% of mothers and 6.8% of fathers), and somatization (13.6% of mothers and 9.7% of fathers; **S1 Table**). Differences between mothers' and fathers' frequencies were not significant (**S1 Table**).

Objective 2: Description of perceived impact of cancer on the couple

We found that mothers and fathers did not differ on their perception of relationship changes following cancer ($F(7, 96) = .96$, $p = .47$; **Table 2**). Inspection of the ICC revealed that the levels of agreement between parents on the perceived impact of the illness ranged from poor to good agreement. Poor agreement was found on time spent together and activities, whereas fair agreement levels were found on intimacy, quality of partner support and conflict. Mothers and fathers reported good agreement on sexuality,

relationship satisfaction, and the overall perceived impact of the illness on the couple (**Table 2**). The vast majority of parents (>75% of mothers and fathers) reported a positive impact on the quality of support with their partner, and more than 50% reported a positive impact on their relationship satisfaction (**S2 Table**). Roughly half of the parents reported that the illness had no significant impact on relationship conflict, whereas some parents reported that their child's illness had a predominately negative effect on their relationship. For instance, a significantly greater proportion of mothers (65%) compared to fathers (43.7%) reported a negative impact of their child's cancer on their level of intimacy with their partner (Wilcoxon z = - 2.861, p = .004; **S2 Table**). These negative changes on intimacy disappeared immediately when their child's treatments were completed or faded with time for most mothers (94%) and fathers (97.7%). A similar trend was observed for other negatively impacted relationship dimensions (**S2 Table**). Approximately half of parents reported a negative impact on the time and activities with their partner, and the majority of them reported that these negative effects either disappeared immediately or faded with time (94.7% of mothers and 98% of fathers). Also, 68% of mothers and 57% of fathers reported that their child's illness had a negative impact on their sexuality as a couple. Among these parents reporting negative effects on sexuality, 8.6% of mothers and 16.9% of fathers reported that the effects were permanent. Altogether, approximately three quarters of parents reported that the period in which their child was ill and treated for leukemia brought them closer together and strengthened their relationship (**Fig 1**). Additionally, the perceived impact of cancer on some relationship dimensions was associated with time since diagnosis, with the more time having passed, the greater the reported positive changes on the 1 to 7 Likert-type scale. Associations

between time since diagnosis and perceived relationship changes were more pronounced among fathers than mothers (**S3 Table**).

Objective 3: Dyadic models for long-term adjustment

Standardized regression coefficients from APIM models for all significant actor, partner, and gender effects are presented in **Table 3**. For both mothers and fathers, no relationship dimensions from the perceived impact of cancer were associated with their global distress. For mothers, greater current relationship satisfaction was associated with them perceiving several positive changes in their relationship with their partner following the illness (**Fig 2**), specifically on: intimacy, quality of partner support, sexuality, relationship satisfaction, and the overall impact of illness on the couple (actor effects). Partner effects for mothers were not statistically significant ($p > .05$). As for psychological distress, mothers reporting that the period of their child's illness brought them closer and strengthened their relationship with their partner was associated with them self-reporting more current anxiety symptoms (actor effect). Moreover, the more positive changes they perceived that the child's illness had on their intimacy with their partner, the fewer depression symptoms that mothers reported (actor effect).

For fathers, their perceived relationship changes following cancer treatments were not significantly associated with their own current relationship satisfaction (i.e., no significant actor effects). Instead, fathers' reported more relationship satisfaction when their partner's reported positive changes on: quality of partner support, conflict, relationship satisfaction, and the overall impact of illness on the couple (partner effects). In terms of psychological distress, fathers reporting that the period of the child's illness brought them closer to their partner and strengthened their relationship were associated

with them reporting more current depression and somatization symptoms (actor effects). Moreover, when mothers reported that the illness had a primarily positive effect on the time and activities with their partner, fathers tended to report more current somatization symptoms (partner effect).

Finally, we found three significant gender differences (**Table 3**). First, when examining the association between current relationship satisfaction and the perceived changes in relationship satisfaction, we found a significant actor effect for mothers but this was not the case for fathers. Second, we found a significant partner effect for predicting fathers' current relationship satisfaction from their partner's perceived changes in relationship satisfaction. The opposite partner effect was not significant. Finally, the actor effect for mothers that related their current depression symptoms and their perceived impact of cancer on intimacy was significant, but this same association was not significant in fathers.

Discussion

In an innovative follow-up study of a hundred and three couples of parents of long-term childhood ALL survivors, we found that only a small subset of parents' reported clinical distress five or more years following their child's leukemia diagnosis. Prevalence of clinical levels of distress was lowest on psychological distress and highest on relationship distress. When exploring relationships among couples, partners tended to agree on the nature of relationship changes (positive, negative or no change) that were experienced following their child's leukemia diagnosis. Within our study, we found that couples' agreements were highest on sexuality as well as general aspects of relationship functioning (e.g., relationship satisfaction) and lowest on psychological distress

symptoms (e.g., anxiety). This finding may indicate that psychological distress is a unique experience for each partner, whereas relationship functioning is a communal and fairly similar experience for both partners.

Description of long-term adjustment

First, we found that 2.9 to 21.4% of parents reported clinical levels of psychological or relationship distress five years or more years following the leukemia diagnosis. This range of clinical distress found in our sample largely resembles the clinical distress range found in a recent review of parents of childhood cancer survivors (compared to 8.8-23% of parents; Ljungman et al., 2014). The proportion of parents that reported clinical levels of anxiety (5.8% of mothers and 6.8% of fathers) and depression symptoms (2.9% of mothers and 6.8% of fathers) in our study also largely resembles the proportions reported by another recent cross-sectional study on long-term acute lymphoblastic leukemia (ALL) survivors and their parents (compared to 7.1% of parents with clinical anxiety and 3.1% of parents with clinical depression; Malpert et al., 2015). A past report examined gender differences in somatization symptoms of parents (164 couples) of children with cancer who were actively being treated or had completed cancer treatments, where mothers reported significantly higher levels on all forms of psychological distress (anxiety, depression, somatization, and global stress) than fathers (Yeh, 2002).

Our current study is the first to examine parents' long-term somatization symptoms specifically during the survivorship period. Likewise, our findings suggest that clinical levels of somatization symptoms also affect a subgroup of parents (13.6% of mothers and 9.7% of fathers). Perhaps the intense stresses associated with the

survivorship period and their child's follow-up appointments manifest themselves in somatic symptoms (e.g., nausea, feeling faint). This finding could also be explained by the fact that these symptoms are often comorbid with anxiety and depression symptoms (Haug, Mykletun, & Dahl, 2004; Löwe et al., 2008). Thus, this implies that physical complaints may be rooted in distress regarding health issues. As for clinical relationship distress, the proportion of parents meeting this threshold in our survivorship study (21.4% of mothers and 20.4% of fathers) is fairly similar to the proportions of parents meeting this threshold at diagnosis (25.5% of mothers and 21.3% of fathers; Burns et al., 2016). However, in that same longitudinal study, 36.2% of mothers and 42.6% of fathers reported clinically significant relationship distress two years later (Burns et al., 2016). Together this longitudinal study and the present study suggest that perhaps particularities of the illness trajectory influence the couples' relationship adjustment, with greater relationship distress two years post diagnosis actually reflecting the stress that can accompany the end of the child's ALL treatments and the re-entry period which is often perceived as lacking support and specialized resources (Muskat et al., 2017). In this qualitative report, parents discussed their fear of relapse, difficulty in adjusting to their life following cancer, and desire for continued support from the health care team (Muskat et al., 2017). Furthermore, a multicenter cross-sectional study found that parents reported feeling the most emotionally connected to their partner at the time of their child's diagnosis and the least emotionally connected to their partner at the beginning and end of treatment (Wiener et al., 2016). Thus, parents' worries regarding relapse and late effects paired with their emotional disconnect with their partner at the end of their child's cancer

treatment appear to be important factors involved in their psychological status and relationship distress.

Next, we found that fathers reported significantly more depression symptoms, but not significantly more frequent clinical levels of depression, than mothers several years after their child's diagnosis. There were no other significant differences between mothers' and fathers' reports of global distress, anxiety, and somatization symptoms. This contrasts with the findings reported by several groups on parental distress during treatment, which indicate that mothers report significantly greater psychological distress (Clarke, McCarthy, Downie, Ashley, & Anderson, 2009; Sultan et al., 2016). It is possible that mothers' distress during treatment subsides with time so that their heightened distress is no longer apparent years after treatment, as was the case in our study. This proposition on the temporal nature of distress is coherent with findings from a longitudinal study on parents' emotional functioning, which suggests that mothers' distress levels largely resemble fathers' distress levels once treatment has ended (Maurice- Stam, Oort, Last, & Grootenhuis, 2008). Time elapsed since diagnosis has also been commonly reported as a significant factor associated with parental distress, both when the child with cancer is on active treatment and in the years following the end of their treatment (Boman, Lindahl, & Björk, 2003). A cross-sectional study of 264 parents (mothers and fathers) of children with cancer at 4 weeks to 14 years post-diagnosis found that the time elapsed since diagnosis explained 2.2 to 13.9% of the variation in aspects of parents' distress, with longer periods since diagnosis being associated with lower levels of distress (Boman et al., 2003).

Description of perceived impact of cancer on couple

We found that couples differed with respect to their relationship adjustment, with some parents reporting primarily negative changes following the illness and others reporting positive changes, which is also coherent with findings from previous studies and reviews (da Silva et al., 2010; Lavee, 2005; Lavee & Mey-Dan, 2003; Silva-Rodrigues, Pan, Sposito, de Andrade Alvarenga, & Nascimento, 2016). Generally, we found that mothers and fathers did not differ on their perceptions of relationship changes following cancer. Most parents tended to perceive that the illness period strengthened their relationship, suggesting that globally, the illness did not undermine their relationship as a couple, despite their perception that some specific areas of their relationship were negatively impacted by the illness. Parents also tended to report positive changes on their relationship satisfaction and the quality of support provided by their partner, as was found in previous reports (da Silva et al., 2010; Lavee & Mey-Dan, 2003; Silva-Rodrigues et al., 2016; Van Schoors et al., 2016; Wiener et al., 2016). The cancer experience itself might have brought them closer to jointly cope with the crisis as a team (Silva-Rodrigues et al., 2016). This sense of marital unity or conjugal resilience has also been qualitatively described as 'we-ness' in recent studies in the field of pediatric cancer (Martin et al., 2014; Martin, Péloquin, Vachon, Duval, & Sultan, 2016).

Nevertheless, the majority of mothers and fathers in our study reported negative changes on aspects of their sexuality, intimacy, and time and activities with their partner. As the child's illness takes precedence over the parents' relationship as a couple, their sexuality often gets pushed aside (da Silva et al., 2010; Lavee & Mey-Dan, 2003; Van Schoors et al., 2016; Wiener et al., 2016). Previous studies have not specifically

examined changes in intimacy and the time spent with their partner and their activities done together. The negative effects that were reported on these two dimensions in our study could be explained by the fact that the child's illness often requires parents' to rearrange family responsibilities, with one parent being the primary caregiver for the ill child at the hospital and the other being responsible for the finances and rest of the family (Goldbeck, 2001; Silva-Rodrigues et al., 2016). This division of labour could lead them to feel distant and less emotionally connected to each other. As for the negative impact on parents' time and activities done with their partner, this could simply be explained by the fact that parents have less time for leisure activities and again illness-related responsibilities take precedence over spending alone time with one's partner. Nonetheless, given that most parents reported that negative effects disappeared with time, this suggests that the experience of having one's child diagnosed and treated for leukemia was not permanently detrimental for the couple and instead was more of a transient challenge that can be overcome with time. This may also explain their overall perception that the period of their child's illness brought them closer together and strengthened their relationship, as mentioned above.

Additionally, we found that the more time that had passed since the child's diagnosis, the greater the likelihood that parents perceived their relationship changes as being more positive in nature, especially for fathers. This is conceptually coherent with the psychological adaptation process (Lazarus & Folkman, 1984; Park, 2010), precisely since survivors were all in remission at the time of this study. It is important to note that the current relationship functioning of parents of ALL survivors in our sample may have biased their recollection of the dynamics of their relationship with their partner during

their child's treatment (Blome & Augustin, 2015; Hensler, Katz, Wiener, Berkow, & Madan-Swain, 2013; Wilson & Ross, 2003). As most parents reported high current relationship satisfaction in our study, this may have tainted their recollection of past events, especially since for some parents the end of their child's cancer treatments were up to 25 years ago.

Impact of cancer on the relationship and long-term adjustment

The current study is the first to examine dyadic associations between mothers and fathers' adjustment to their child's cancer in the survivorship period using predictors of changes in relationship dynamics within couples as a result of the cancer experience. We found that mothers' adjustment was exclusively self-related (actor effects), whereas fathers' adjustment was mostly partner-related (partner effects) and to a lesser extent self-related. These results are both coherent with and extend results from previous studies in this field. Similar trends in actor and partner effects for mothers and fathers were reported in a recent dyadic, longitudinal study of parents of children with cancer in the first two years following their diagnosis (Burns et al., 2016). That is, mothers' marital adjustment 2 years later was explained by their own perceived family functioning at diagnosis (actor effects), whereas fathers' marital adjustment was explained by their own mood and family functioning (actor effects), as well as by their partner's family functioning at diagnosis (partner effects) (Burns et al., 2016).

Predicting relationship satisfaction

Parents of children with cancer are often well adjusted in their relationship with their partner (Van Schoors et al., 2016), and this finding on general relationship functioning (e.g., relationship satisfaction) is consistent with what was found in our

study. In the present study we found that mothers' current relationship satisfaction was associated with their own perceptions of positive relationship changes (intimacy, partner support, sexuality, satisfaction, and overall impact of the illness), whereas fathers' current relationship satisfaction was associated with their partner's perceived positive relationship changes (partner support, conflict, satisfaction, and overall impact of the illness). These findings seem to suggest that long-term relationship adjustment is an independent experience for mothers, but an interdependent experience for fathers. In this way, fathers' relationship satisfaction might be at least partially dependent on how their partner views their relationship and the subsequent changes in their relationship dynamics following their child's illness.

It is possible that this differential pattern in mothers and fathers would be due to their fundamental differences in support-seeking behaviours. Several studies have suggested that while mothers of children with cancer receive their social support from various sources (including their partner, friends, family, and the health care team), fathers primarily seek support from their partner (Lavee, 2005; Leventhal-Belfer, Bakker, & Russo, 1993). Support-seeking has been found to be a more frequent coping strategy among mothers than fathers even after the child's cancer treatments have ended (Norberg, Lindblad, & Boman, 2006). In fact, a cross-sectional study of parents of children with cancer (N = 35 couples) found that while mothers' reliance on social support lowered their distress, fathers felt less distressed when their relationship with their partner was strong (Lavee, 2005). This tendency for fathers to largely depend on their partner for support, could in turn explain why it is that mothers' experiences during the child's illness have such a substantial impact on fathers even several years after their

child's diagnosis. It is also possible that since mothers are often the primary caregivers for the ill child and more frequently the ones at the hospital, fathers use mothers' experiences as a bridge for understanding their child's cancer experience and its impact on the family. Perhaps mothers are also more emotionally expressive about their perceptions on changes in relationship dynamics following their child's diagnosis, and that fathers use their partner's perceptions as a gauge for their current relationship functioning. For instance, if mothers express significant appreciation towards their partner's support during the illness then perhaps fathers will interpret this as an indication of the strength of their relationship and in turn report greater relationship satisfaction even several years later. Consistently, we found that the more positive the perceived cancer-related effects on the relationship, the greater parents' reports of current relationship satisfaction. In other words, positive relationship experiences during treatment appear to translate into positive relationship experiences during the survivorship period.

Predicting psychological distress

With the current study, we found that changes that occurred in couples' relationships during their child's leukemia treatments were significantly related to both partner's psychological distress during the survivorship period. Specifically, mothers' perceiving that the illness had a positive effect on their intimacy and that it strengthened their relationship with their partner was associated with them reporting less depression and more anxiety symptoms at the time of our follow-up study. Effects were also found for fathers, whereby their perception that the illness period brought them closer and strengthened their relationship was associated with them reporting greater current

depression and somatization symptoms. We also found that certain relationship changes perceived by mothers (time and activities) were associated with fathers reporting greater current psychological distress (somatization symptoms). At first glance these findings seem counterintuitive, as we would expect that recalling positive relationship changes during the child's treatment would be associated with less distress later. Yet, given that this study was a cohort recall aimed at examining long-term adverse effects in ALL survivors, the very nature of this study could explain the present phenomenon. Since parents were asked to self-report psychological distress in the last week including on the day of their child's follow up testing, parents' heightened anxiety, depression, and somatization symptoms are understandable and thus may not be a representative depiction of their standard level of distress (e.g., trait anxiety). Instead it might reflect state-dependent distress, which is mostly related to their illness-related concerns regarding late effects and limitations that could be found in their child's upcoming medical follow-up appointment. Furthermore, it is possible that self-reflecting on their child's illness in the days leading up to the follow-up study, even when related to positive relationship effects, could have served as an inadvertent mood prime and bias for their subsequent reports of their psychological status.

Given the correlational nature of our study, greater current psychological distress was associated with recalling more positive relationship changes during the illness (including feeling that the illness period strengthened their relationship), and less current psychological distress was associated with recalling more negative relationship changes during the illness (including feeling that the illness period was detrimental to their relationship). This seems to suggest two distinct profiles of parents, those who reported

positive relationship changes-high distress and those who reported *negative relationship changes-low distress*. In this way, it appears that perhaps the presence of (or ability to report) difficult relationship dynamics during the illness might be more adaptive long-term in reducing parents' psychological distress in the survivorship period. One other possible explanation for this is that these profiles actually reflect gender roles in parenting. Even today, mothers of children with cancer are typically the primary caregivers for the ill child, accompanying them at the hospital and often taking a leave of absence from work or reducing their hours considerably in order to care for the ill child (Chesler & Parry, 2001; Limburg, Shaw, & McBride, 2008; Mader et al., 2016). Besides maintaining employment during the child's illness, fathers often take on additional family responsibilities such as caring for the other children and doing household chores (Chesler & Parry, 2001). Fathers often feel isolated and left out of the child's treatment decisions (Chesler & Parry, 2001), and just because they are often at work this does not mean that they are not constantly thinking about their ill child. As a result, fathers of children with cancer report considerable gender role conflict (Chesler & Parry, 2001; Hall, 2010). This description reflects traditional gender roles endorsed by parents during their child's cancer treatment. Following the end of their child's cancer treatments, parents, especially mothers, can have difficulty returning to work due to career setbacks or missed promotions (Limburg et al., 2008). In fact, mothers were more likely to stop working altogether after their child's cancer diagnosis (Limburg et al., 2008; Lindahl Norberg, Montgomery, Bottai, Heyman, & Hovén, 2017). Compared to control parents in the general population, mothers of cancer survivors are more commonly not employed while fathers of cancer survivors are more commonly employed full-time (Mader et al., 2016).

It appears near to impossible for parents to return to the status quo of what their life was before cancer. By the time that their child is in the survivorship phase, parents' caregiving responsibilities have tapered off considerably, but their financial well-being is often still an ever-present concern for the couple. The couples' earlier decision to take on traditional gender roles might have lead them to report more positive relationship dynamics during the illness, but this decision might have also led to more financial difficulties and long-term distress for the couple later on.

On the contrary, couples that took on non-traditional gender roles, with both parents alternating between caring for the ill child and working, might have experienced more relationship difficulties during the illness (thus leading partners to feel that this period distanced them) but less psychological distress later. Alternating between caregiving and financial responsibilities, would allow fathers to be more present for the ill child at the hospital and allow mothers to not exclusively put their career in jeopardy. Such restructuring of the family unit to endorse this non-traditional division of responsibilities will undoubtedly come with its own difficulties. For example, if one partner is not content with this arrangement this can negatively bias how they view their relationship and the impact of their child's illness on their relationship. However, in the long-term these couples might be better able to adjust to their life after cancer. With our study, at the time of follow-up (notably during the survivorship period) a higher proportion of fathers (76.7%) were employed full-time than mothers (63.1%). However, the proposed couples' profiles could not be tested since division of labour and caregiving responsibilities were not assessed. Future studies should formally test this proposed

association, linking cancer-related relationships changes and long-term psychological adjustment in couples.

Limitations

Although the current study follows a relatively large cohort of parents whose children were treated for ALL for a long period of time, its cross-sectional nature prevents us from interpreting associations as causal links. For example, causality between perceptions of the impact of cancer on couples' relationship and parents' adjustment can only be hypothesized, as it is possible that retrospective evaluations of the impact would be influenced by their current psychological status. Furthermore, we should be mindful that our sample only reflects the experiences of relatively stable couples that were together both during their child's illness and at the time of follow-up. This restrictive definition of couples excludes all mothers and fathers who have separated from the other parent and precludes including couples from reconstituted families (i.e., step-parents). Next, the homogeneity of diagnosis in this sample facilitates comparisons of the long-term effects and adjustment of survivors and their parents. However, since rates of survival for childhood ALL are relatively high, the reports of parents in this sample might not reflect the experiences of parents of survivors with more sombre survival expectations or morbidities, such as parents of children treated for brain tumour or those who had relapsed. Finally, given the final participation rate we cannot rule out the effect of possible selection bias. Although we tried to include all parents of children treated with standard DFCI protocols from 1987 to 2005, those who did not respond or send back their questionnaires may be those that are less well-adjusted, as evidenced with greater psychological distress or more substantial relationship challenges.

Conclusion

In our cross-sectional study of the retrospective adjustment experiences of 103 couples of parents of childhood ALL survivors, we found that parents were generally well-adjusted, with only a small subset (2.9-21.4%) reporting clinical distress at follow-up. Using an interdependence model, this study was the first to examine the dyadic adjustment and relationship change experiences of parents of childhood ALL survivors. In doing so, we found that mothers' adjustment (relationship satisfaction and psychological distress) was solely associated with her own perceptions of changes in relationship dynamics, while fathers' adjustment was associated with both their own perceptions and those of their partner. These findings suggest that couples' adjustment is an interdependent process. It is thus possible that by strengthening dimensions of relationship functioning among mothers, we would actually be fostering better adjustment for both parents. This observation could be translated into new integrated and family-based approaches for addressing individual and interpersonal distress among parents. For instance, providing a couples-based support program, which uses what other researchers have referred to as 'relationship talk' (i.e., partners discussing their relationship and dimensions within that relationship; Badr & Acitelli, 2005) or 'social sharing' (i.e., expressing their thoughts and feelings regarding cancer; Boinon et al., 2014) during the child's cancer treatments, could help both partners address their interpersonal difficulties early in the illness trajectory. This would also allow mothers to openly address their perceptions of negative relationship changes with their partner. This in turn could help to foster stronger general relationship functioning for both partners and promote relationship satisfaction in this vulnerable population.

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Table 1. *Parents' and childhood ALL survivors' characteristics (n = 103).*

Table 1. Parents' and childhood ALL survivors' characteristics (n = 103)

		Couples M (SD) or N (%)	
Parents' characteristics	**Mothers**		**Fathers**
Length of relationship, years		29.90 (7.63)	
Age at diagnosis, years	35.76 (5.83)		37.70 (5.12)
Age at follow-up interview, years	51.56 (6.75)		53.63 (6.11)
Highest education level			
High school	21 (20.4)		37 (35.9)
Undergraduate	50 (48.5)		46 (44.7)
Graduate	12 (11.7)		7 (6.8)
Other (e.g., high school not completed)	20 (19.4)		13 (12.6)
Primary occupation			
Working, full-time	65 (63.1)		79 (76.7)
Working, part-time	12 (11.7)		4 (3.9)
Other (e.g., retired, unemployed, at home)	26 (25.2)		20 (19.4)
Financial income (gross, $CAD)			
< $49,999	67 (65.0)		30 (29.1)
$50,000 - 89,999	30 (29.1)		42 (40.8)
$90,000 +	6 (5.8)		31 (30.1)
Language			
French	98 (95.1)		97 (94.2)
English	0 (0)		1 (1.0)
Other	5 (4.9)		5 (4.9)

Survivors' characteristics	Couples M (SD) or N (%)
Age at diagnosis, years	6.26 (4.78)
Age at follow-up interview, years	22.09 (6.66)
Child (≤ 18)	41 (39.8)
Adolescent/young adult (19-25)	35 (34.0)
Adult (≥ 26)	27 (26.2)
Time since diagnosis, years	15.46 (5.12)
Range	5-27
Time since end of treatment, years	13.28 (5.20)
Range	3-25
Sex	
Female	59 (57.3)
Male	44 (42.7)
ALL relapse risk group	
Standard risk	45 (44.1)
High risk	57 (55.9)
Treatment protocol	
DFCI 87-01	10 (9.7)
DFCI 91-01	19 (18.4)
DFCI 95-01	34 (33.0)
DFCI 2000-01	32 (31.1)
DFCI 2005-01	8 (7.8)
Radiotherapy	
No	39 (37.9)
Yes	64 (62.1)
Known long-term complications	
No	45 (44.1)
Yes	57 (55.9)
Relationship status	
Single	73 (70.9)
Married	4 (3.9)
Divorced	2 (1.9)
Common law partner	24 (23.3)
Highest education level	
High school not yet completed	34 (33.0)
High school	19 (18.4)
Undergraduate	37 (35.9)
Graduate	1 (1.0)
Other (e.g., vocational diploma)	12 (11.7)
Financial income (gross, $CAD)	
< $49,999	87 (84.5)
$50,000 - 89,999	15 (14.6)
$90,000 +	1 (1.0)
Primary occupation	
Working, full-time	39 (37.9)
Working, part-time	30 (29.1)
Other (e.g., student, unpaid work, unemployed)	34 (33.0)
Language	
French	98 (95.1)
English	2 (1.9)
Other	3 (2.9)

Table 2. *Description of the perceived impact of cancer, and psychological and relationship adjustment in a sample of 103 couples of parents whose children were treated for ALL (n = 103).*

	Couples				
	Mothers *M (SD)*	**Fathers** *M (SD)*	**Repeated-measures MANOVA testing gender differences**	**ICC (95% CI)**	**Levels of agreement**
Impact of Cancer on the Couple					
Intimacy	3.55 (1.74)	3.94 (1.76)	$F(1,102) = 3.768$, $p = .055$	.487, $p = .000$ (.247-.651)	Fair
Quality of partner support	5.80 (1.43)	5.81 (1.19)	$F(1,102) = .086$, $p = .770$	.480, $p = .001$ (.230-.649)	Fair
Sexuality	3.17 (1.46)	3.20 (1.38)	$F(1,102) = .079$, $p = .779$	.683, $p = .000$ (.531-.785)	Good
Conflict	4.02 (1.16)	4.02 (1.11)	$F(1,102) = .000$, $p = 1.000$	.539, $p = .000$ (.317-.688)	Fair
Time & activities	3.55 (1.56)	3.66 (1.45)	$F(1,102) = .322$, $p = .571$	.329, $p = .023$ (.007-.547)	Poor
Relationship satisfaction	4.69 (1.75)	4.69 (1.46)	$F(1,102) = .000$, $p = 1.000$	.636, $p = .000$ (.462-.754)	Good
Overall impact on couple	5.49 (1.80)	5.58 (1.62)	$F(1,102) = .360$, $p = .550$	.703, $p = .000$ (.561-.799)	Good
Dyadic Adjustment Scale (DAS-4)					
Relationship satisfaction	15.36 (3.69)	15.55 (3.50)	$F(1,102) = .441$, $p = .508$	.796, $p = .000$ (.699-.862)	Excellent
Brief Symptom Inventory (BSI-18)					
Global Symptom Index (GSI)	46.11 (9.41)	46.39 (9.67)	$F(1,102) = .058$, $p = .810$	.251, $p = .075$ (-.111-.494)	Poor
Anxiety	47.38 (8.91)	45.56 (9.26)	$F(1,102) = 2.839$, $p = .095$	.070, $p = .354$ (-.362-.367)	Poor
Depression	**44.75 (7.33)**	**46.25 (7.66)**	$F(1,102) = 3.986$, $p = .049$	.212, $p = .110$ (-.151-.462)	Poor
Somatization	49.24 (8.39)	48.90 (8.43)	$F(1,102) = .100$, $p = .753$	.376, $p = .009$ (.076-.578)	Poor

Note. Means and standard deviations are computed using t-scores on the BSI-18. Bolded means indicate a significant gender difference. To facilitate interpretation, untransformed means and standard deviations are presented here.

Table 3. *Actor, partner, and gender effects as identified by APIM models predicting relationship satisfaction and psychological distress from the perceived impact of cancer on the couple (n = 100).*

	Actor effect			Partner effect		
Predictors of relationship satisfaction	**Mother**	**Father**	**Gender difference**	**Mother**	**Father**	**Gender difference**
Impact of Cancer on Couple						
Intimacy	β = .278*		p = .237			
Quality of partner support	β = .337**		p = .095		β = .201*	p = .405
Sexuality	β = .293*		p = .307			
Conflict					β = .197*	p = .977
Relationship satisfaction	β = .559**		p = .027		β = .288**	p = .028
Overall impact of illness	β = .453**		p = .141		β = .260*	p = .334
Predictors of psychological distress						
Anxiety						
Overall impact of illness	β = .272*		p = .766			
Depression						
Intimacy	β = -.236*		p = .009			
Overall impact of illness		β = .238*	p = .977			
Somatization						
Time & activities					β = .190*	p = .280
Overall impact of illness		β = .273*	p = .195			

Note. All possible associations were tested. For clarity, only significant regression coefficients are displayed (*p < .05, **p < .01). Full results are available from the authors.

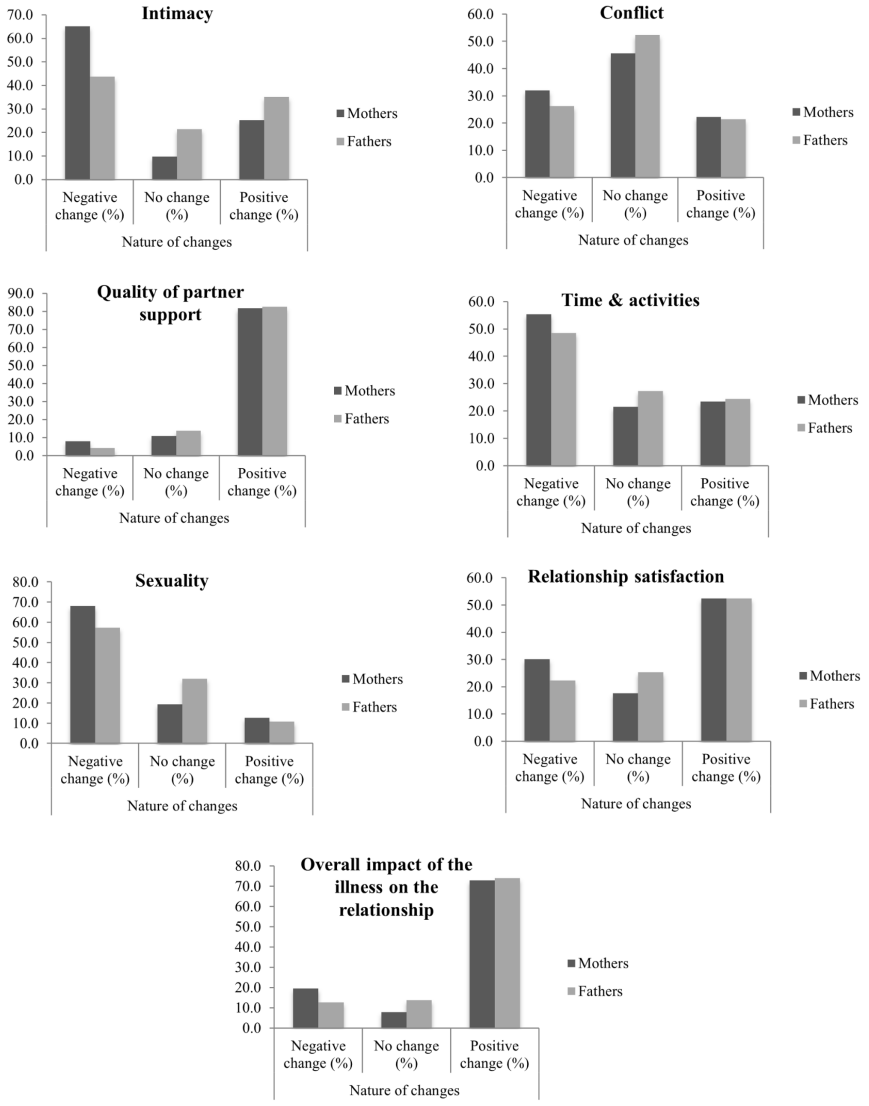


Figure 1. *Bar charts displaying the nature of relationship changes for mothers and fathers (n =103).*

Note. Relationship dimensions are represented on a 1 to 7 Likert-type scale, whereby participants' scores are classified into: negative change (scores of 1-3), no change (scores of 4), or positive change (scores of 5-7).

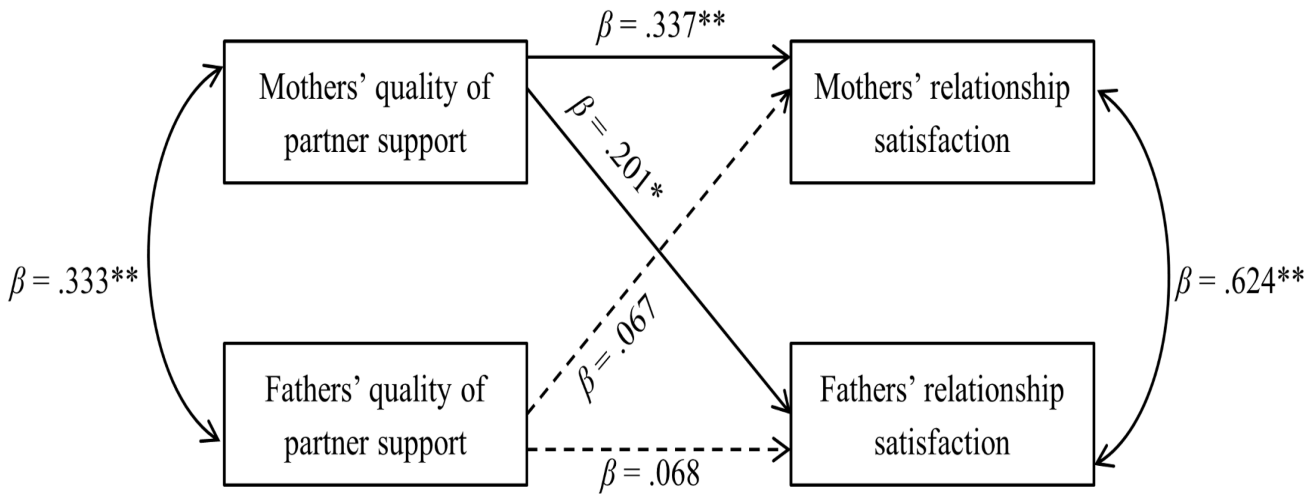
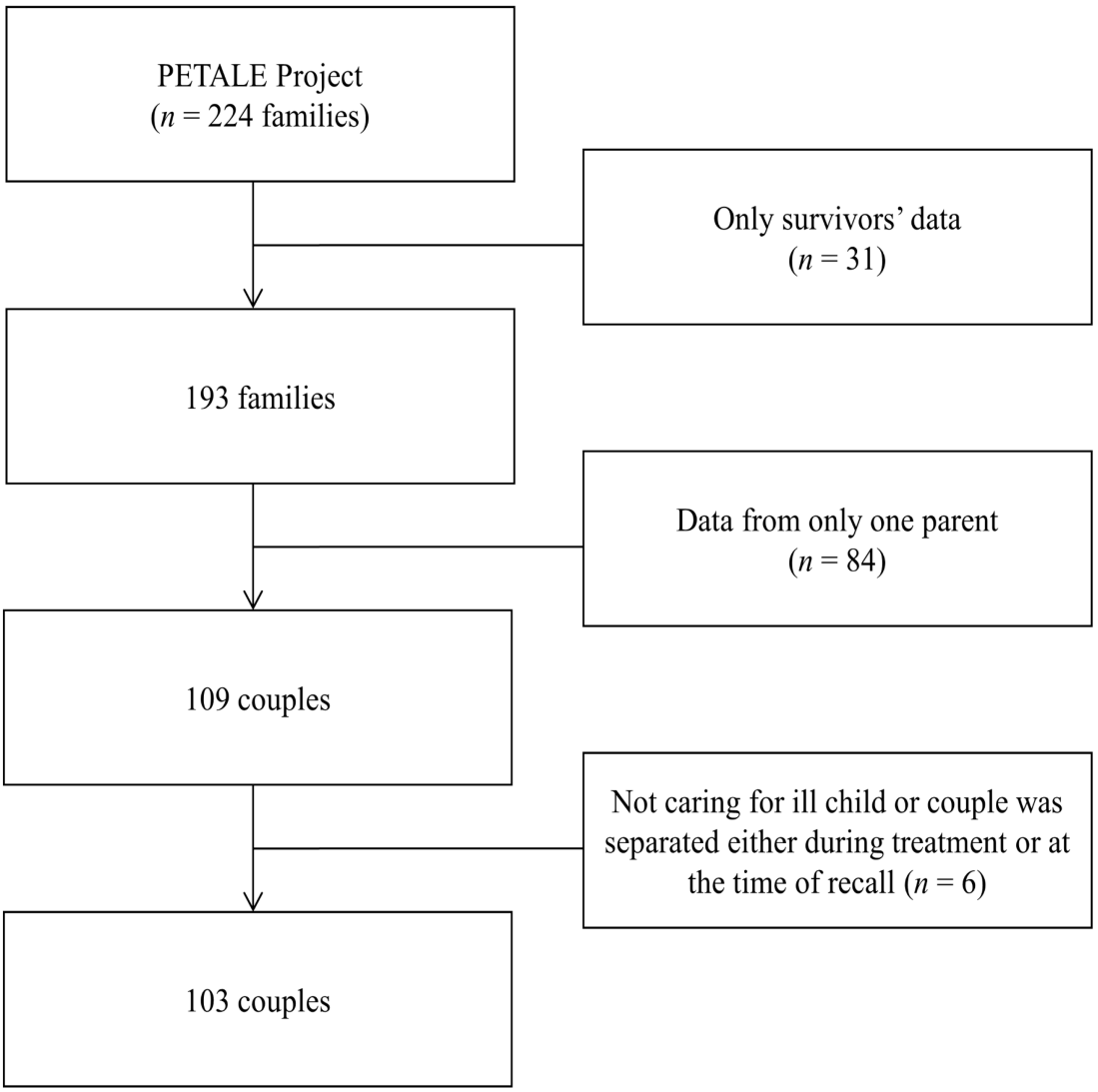


Figure 2. *Actor-partner interdependence model (APIM) predicting current relationship satisfaction from perceived changes in quality of partner support (n = 100 couples)*

*Note. ** p < .01, * p < .05; the dashed lines represent non-significant associations.*



S1 Figure . *Flowchart of study participants.*

Note. Depression variables were severely skewed, and parents in three couples had extreme depression scores (z score > 3.5). Data from these three couples were retained in all analyses for Objectives 1 and 2, but due to normality concerns they were excluded from the dyadic analyses in Objective 3.

S1 Table. *Proportion of mothers and fathers scoring within the clinical range (i.e., positive caseness) on adjustment variables (n = 103).*

	Couples				
	Mothers		Fathers		
Individuals within clinical range	***n***	**%**	***n***	**%**	**McNemar tests**
Dyadic Adjustment Scale (DAS-4)					
Relationship satisfaction	22	21.4%	21	20.4%	$p = 1.000$
Brief Symptom Inventory (BSI-18)					
Global Symptom Index	7	6.8%	8	7.8%	$p = 1.000$
Anxiety	6	5.8%	7	6.8%	$p = 1.000$
Depression	3	2.9%	7	6.8%	$p = .344$
Somatization	14	13.6%	10	9.7%	$p = .503$

S2 Table. *Proportion of parents that reported negative change, no change and positive change in relationship dimensions on the Impact of Cancer on the Couple (n =103).*

	Couples						
	Mothers			Fathers			
Relationship Dimensions	Nature of changes			Nature of changes			
Duration of the negative change	**Negative change (%)**	**No change (%)**	**Positive change (%)**	**Negative change (%)**	**No change (%)**	**Positive change (%)**	**Wilcoxon tests**
Intimacy	65.0	9.7	25.2	43.7	21.4	35.0	***z* = - 2.861, *p* = .004**
Disappeared immediately (%)	31.3			33.3			
Faded with time (%)	62.7			64.4			
Permanent (%)	6.0			2.2			
Quality of partner support	7.8	10.7	81.6	3.9	13.6	82.5	*z* = - .932, *p* = .351
Disappeared immediately (%)	25.0			50.0			
Faded with time (%)	62.5			25.0			
Permanent (%)	12.5			25.0			
Sexuality	68.0	19.4	12.6	57.3	32.0	10.7	*z* = -1.110, *p* = .267
Disappeared immediately (%)	32.9			39.0			
Faded with time (%)	58.6			44.1			
Permanent (%)	8.6			16.9			
Conflict	32.0	45.6	22.3	26.2	52.4	21.4	*z* = - .642, *p* = .521
Disappeared immediately (%)	33.3			59.3			
Faded with time (%)	60.1			33.3			
Permanent (%)	6.1			7.4			
Time & activities	55.3	21.4	23.3	48.5	27.2	24.3	*z* = -.683, *p* = .495
Disappeared immediately (%)	39.3			54.0			
Faded with time (%)	55.4			44.0			
Permanent (%)	5.4			2.0			
Relationship satisfaction	30.1	17.5	52.4	22.3	25.2	52.4	*z* = - 1.091, *p* = .275
Disappeared immediately (%)	22.6			30.4			
Faded with time (%)	71.0			56.5			
Permanent (%)	6.5			13.0			
Overall impact of illness on relationship	19.4	7.8	72.8	12.6	13.6	73.8	*z* = -1.263, *p* = .207

Note. Relationship dimensions are represented on a 1 - 7 scale. Classifications of scores are as follows: Scores 1 - 3 = negative effect, 4 = no effect, and 5 - 7 = positive effect. Bolded text indicates a significant gender difference.

S3 Table. *Correlations between perceived impact of cancer and adjustment variables in mothers and fathers of children treated for acute lymphoblastic leukemia (n = 103).*

	1	2	3	4	5	6	7	8	9	10	11	12	13
1. Time elapsed since diagnosis		.147	.083	.172	-.023	.205*	.030	.046	-.131	.074	.055	.059	.116
Impact of Illness													
2. Intimacy	.216*	.327**	.423**	.657**	.404**	.570**	.562**	.477**	.283**	-.099	-.035	-.145	-.047
3. Quality of partner support	.121	.286**	.317**	237*	.476**	.275**	.525**	.553**	.369**	.077	.165	.086	.024
4. Sexuality	.257**	.602**	.083	.517**	.213*	.620**	.465**	.342**	.272**	-.059	-.047	-.136	.026
5. Conflict	.109	.432**	.285**	.412**	.366**	.244*	.435**	.534**	.267**	.141	.189	.152	.044
6. Time & activities	.315**	.412**	.133	.457**	.499**	.197*	.354**	.284**	.120	.055	.058	-.008	.106
7. Relationship satisfaction	.154	.469**	.476**	.488**	.450**	.437**	.472**	.616**	.565**	-.083	-.020	-.080	-.085
8. Overall impact of illness	.031	.393**	.500**	.283**	.368**	.319**	.634**	.543**	.511**	.105	.137	.132	.003
Dyadic Adjustment Scale (DAS-4)													
9. Relationship satisfaction	-.007	.149	.140	.206*	.252*	.196*	.333**	.331**	.661**	-.191	-.103	-.236*	-.164
Brief Symptom Inventory (BSI-18)													
10. Global Severity Index (GSI)	-.126	.071	.002	.049	-.049	-.012	.036	.145	-.208*	.142	.869**	.758**	.825**
11. Anxiety	-.184	.142	.034	.092	-.010	.016	.051	.154	-.154	.847**	.037	.593**	.599**
12. Depression	-.112	.139	.015	.089	-.072	-.029	.014	.134	-.181	.826**	.694**	.122	.475**
13. Somatization	-.047	.033	.020	.085	-.007	.031	.103	.171	-.120	.848**	.570**	.620**	.230*

Note. Mothers' bivariate correlations are above the diagonal and fathers' bivariate correlations are below the diagonal. Interrelationships between the partners' variables are displayed on the diagonal. ** $p < .01$, * $p < .05$

S1 File. *Impact du cancer sur le couple.*

IMPACT DU CANCER SUR LE COUPLE

(Péloquin & Sultan, 2013)

1. Quel âge aviez-vous au début de votre relation de couple avec le père / la mère de votre enfant survivant de cancer? _____

2. Depuis combien de temps êtes-vous en couple (ou pendant combien de temps avez-vous été en couple) avec le père / la mère de votre enfant survivant de cancer? _____

3. Quel est votre statut conjugal actuel?
    1. ___ En couple avec le père / la mère de mon enfant survivant de cancer (allez directement à la question 5)
    2. ___ En couple avec un(e) autre conjoint(e) (répondez à la question 4)
    3. ___ Seul(e) (séparé / divorcé) (répondez à la question 4)
    4. ___ Veuf / veuve (allez directement à la question 5)

4. Si vous êtes séparé(e) / divorcé(e) du père / de la mère de votre enfant survivant de cancer, à quel point la maladie de votre enfant a-t-elle contribué à cette séparation selon vous ?

    1. ___ La maladie (et le contexte entourant la maladie) n'est *pas du tout liée* à notre séparation.
    2. ___ La maladie (et le contexte entourant la maladie) a *légèrement* contribué à notre séparation.
    3. ___ La maladie (et le contexte entourant la maladie) a *modérément* contribué à notre séparation.
    4. ___ La maladie (et le contexte entourant la maladie) a *beaucoup* contribué à notre séparation.
    5. ___ La maladie (et le contexte entourant la maladie) est *entièrement responsable* de notre séparation.

5. Étiez-vous en couple avec le père / la mère de votre enfant survivant de cancer au moment où votre enfant était malade et suivi pour des traitements à Sainte-Justine?

    1. ___ Oui, nous étions en couple (répondez à la question 6)
    0. ___ Non, nous étions séparés (ne répondez pas aux questions 6 et 7)

Les questions suivantes traitent de la période pendant laquelle votre enfant était malade et suivi pour des traitements à Ste-Justine. SVP répondre à ces questions en repensant à votre relation de couple pendant cette période.

6. Lorsque les couples vivent des situations de stress important, incluant la maladie d'un enfant, il est possible que ces situations influencent leur relation de couple de diverses façons. À l'aide des échelles de réponse suivantes, veuillez évaluer à quel point la maladie de votre enfant a eu un effet sur les dimensions conjugales suivantes :

6.A.1 L'intimité entre vous et votre conjoint(e) :

1	*2*	*3*	*4*	*5*	*6*	*7*
Effet très négatif	*Effet modérément négatif*	*Effet légèrement négatif*	*Aucun effet*	*Effet légèrement positif*	*Effet modérément positif*	*Effet très positif*

6.A.2. Si vous avez répondu 1, 2 ou 3, ces effets négatifs ont-ils perduré lorsque les traitements de votre enfant ont été terminés ?

1. ___ Les effets négatifs ont disparu immédiatement
2. ___ Les effets négatifs ont perduré, mais se sont estompées avec le temps
3. ___ Les effets négatifs ont été permanents

6.B.1. La qualité du soutien entre vous et votre conjoint(e) :

1	*2*	*3*	*4*	*5*	*6*	*7*
Effet très négatif	*Effet modérément négatif*	*Effet légèrement négatif*	*Aucun effet*	*Effet légèrement positif*	*Effet modérément positif*	*Effet très positif*

6.B.2. Si vous avez répondu 1, 2 ou 3, ces effets négatifs ont-ils perduré lorsque les traitements de votre enfant ont été terminés ?

1. ___ Les effets négatifs ont disparu immédiatement
2. ___ Les effets négatifs ont perduré, mais se sont estompées avec le temps
3. ___ Les effets négatifs ont été permanents

6.C.1. La sexualité entre vous et votre conjoint(e) :

1	*2*	*3*	*4*	*5*	*6*	*7*
Effet très négatif	*Effet modérément négatif*	*Effet légèrement négatif*	*Aucun effet*	*Effet légèrement positif*	*Effet modérément positif*	*Effet très positif*

6.C.2. Si vous avez répondu 1, 2 ou 3, ces effets négatifs ont-ils perduré lorsque les traitements de votre enfant ont été terminés ?

1. ___ Les effets négatifs ont disparu immédiatement
2. ___ Les effets négatifs ont perduré, mais se sont estompées avec le temps
3. ___ Les effets négatifs ont été permanents

6.D.1. Les conflits entre vous et votre conjoint(e) :

1	*2*	*3*	*4*	*5*	*6*	*7*
Effet très négatif	*Effet modérément négatif*	*Effet légèrement négatif*	*Aucun effet*	*Effet légèrement positif*	*Effet modérément positif*	*Effet très positif*

6.D.2. Si vous avez répondu 1, 2 ou 3, ces effets négatifs ont-ils perduré lorsque les traitements de votre enfant ont été terminés ?

1. ___ Les effets négatifs ont disparu immédiatement
2. ___ Les effets négatifs ont perduré, mais se sont estompées avec le temps
3. ___ Les effets négatifs ont été permanents

6.E.1. Le temps passé et les activités réalisées avec votre conjoint(e) :

1	*2*	*3*	*4*	*5*	*6*	*7*
Effet très négatif	*Effet modérément négatif*	*Effet légèrement négatif*	*Aucun effet*	*Effet légèrement positif*	*Effet modérément positif*	*Effet très positif*

6.E.2. Si vous avez répondu 1, 2 ou 3, ces effets négatifs ont-ils perduré lorsque les traitements de votre enfant ont été terminés ?

1. ___ Les effets négatifs ont disparu immédiatement
2. ___ Les effets négatifs ont perduré, mais se sont estompées avec le temps
3. ___ Les effets négatifs ont été permanents

6.F.1. Votre satisfaction conjugale générale :

1	*2*	*3*	*4*	*5*	*6*	*7*
Effet très négatif	*Effet modérément négatif*	*Effet légèrement négatif*	*Aucun effet*	*Effet légèrement positif*	*Effet modérément positif*	*Effet très positif*

6.F.2. Si vous avez répondu 1, 2 ou 3, ces effets négatifs ont-ils perduré lorsque les traitements de votre enfant ont été terminés ?

1. ___ Les effets négatifs ont disparu immédiatement
2. ___ Les effets négatifs ont perduré, mais se sont estompées avec le temps
3. ___ Les effets négatifs ont été permanents

7. De façon générale, comment décririez-vous l'impact de la période pendant laquelle votre enfant était malade sur votre relation de couple ?

1	*2*	*3*	*4*	*5*	*6*	*7*
Cette période nous a éloigné / a été néfaste pour notre relation			*Cette période n'a eu aucun effet sur notre relation*			*Cette période nous a rapproché / a renforcé notre relation*

S2 File. *Impact of Cancer on the Couple.*

IMPACT OF CANCER ON THE COUPLE

(Peloquin & Sultan, 2013)

1. How old were you at the beginning of your relationship with the father/mother of your child cancer survivor ? _____

2. How long have you been a couple (or how for how long were you a couple) with the father/mother of your child cancer survivor ? _____

3. What is your current marital status ?
    1. ___ In a relationship with the father/mother of my child cancer survivor (go directly to question 5)
    2. ___ In a relationship with another partner (answer question 4)
    3. ___ Single (separated/divorced) (answer question 4)
    4. ___ Widowed (go directly to question 5)

4. If you are separated/divorced from the father/mother of your child cancer survivor, in your opinion how much did your child's illness contributed to this separation ?

    1. ___ The illness (and the context surrounding the illness) is *not at all related to* our separation.
    2. ___ The illness (and the context surrounding the illness) *contributed slightly* to our separation.
    3. ___ The illness (and the context surrounding the illness) *moderately contributed* to our separation.
    4. ___ The illness (and the context surrounding the illness) *greatly contributed* to our separation.
    5. ___ The illness (and the context surrounding the illness) is *entirely responsible* for our separation

5. Were you in a relationship with the father / mother of your child cancer survivor <u>at the time that your child was sick and followed for treatment at Sainte-Justine</u>?

    1. ___ Yes, we were a couple (answer question 6)
    0. ___ No, we were separated (do not answer questions 6 and 7)

The following questions deal with the period in which your child was sick and followed for treatment. Please answer these questions, thinking back to your relationship during this period.

6. When couples experience severe stress, including a child's illness, these situations may influence their relationship in a variety of ways. Using the following response scales, please rate <u>the extent to which your child's illness had an effect</u> on the following relationship dimensions:

6.A.1 Intimacy between you and your partner:

1	*2*	*3*	*4*	*5*	*6*	*7*
Very negative effect	*Moderately negative effect*	*Slightly negative effect*	*No effect*	*Slightly positive effect*	*Moderately positive effect*	*Very positive effect*

6.A.2. If you answered 1, 2 or 3, did these negative effects persist when your child's treatments were completed?

1. ___ The negative effects disappeared immediately
2. ___ The negative effects have persisted, but have faded with time
3. ___ The negative effects were permanent

6.B.1. The quality of support between you and your partner:

1	*2*	*3*	*4*	*5*	*6*	*7*
Very negative effect	*Moderately negative effect*	*Slightly negative effect*	*No effect*	*Slightly positive effect*	*Moderately positive effect*	*Very positive effect*

6.B.2. If you answered 1, 2 or 3, did these negative effects persist when your child's treatments were completed?

1. ___ The negative effects disappeared immediately
2. ___ The negative effects have persisted, but have faded with time
3. ___ The negative effects were permanent

6.C.1. Sexuality between you and your partner:

1	*2*	*3*	*4*	*5*	*6*	*7*
Very negative effect	*Moderately negative effect*	*Slightly negative effect*	*No effect*	*Slightly positive effect*	*Moderately positive effect*	*Very positive effect*

6.C.2. If you answered 1, 2 or 3, did these negative effects persist when your child's treatments were completed?

1. ___ The negative effects disappeared immediately
2. ___ The negative effects have persisted, but have faded with time
3. ___ The negative effects were permanent

6.D.1. Conflicts between you and your partner:

1	*2*	*3*	*4*	*5*	*6*	*7*
Very negative effect	*Moderately negative effect*	*Slightly negative effect*	*No effect*	*Slightly positive effect*	*Moderately positive effect*	*Very positive effect*

6.D.2. If you answered 1, 2 or 3, did these negative effects persist when your child's treatments were completed?

1. ___ The negative effects disappeared immediately
2. ___ The negative effects have persisted, but have faded with time
3. ___ The negative effects were permanent

6.E.1. Time spent and activities done with your partner:

1	*2*	*3*	*4*	*5*	*6*	*7*
Very negative effect	*Moderately negative effect*	*Slightly negative effect*	*No effect*	*Slightly positive effect*	*Moderately positive effect*	*Very positive effect*

6.E.2. If you answered 1, 2 or 3, did these negative effects persist when your child's treatments were completed?

1. ___ The negative effects disappeared immediately
2. ___ The negative effects have persisted, but have faded with time
3. ___ The negative effects were permanent

6.F.1. Your overall relationship satisfaction:

1	*2*	*3*	*4*	*5*	*6*	*7*
Very negative effect	*Moderately negative effect*	*Slightly negative effect*	*No effect*	*Slightly positive effect*	*Moderately positive effect*	*Very positive effect*

6.F.2. If you answered 1, 2 or 3, did these negative effects persist when your child's treatments were completed?

1. ___ The negative effects disappeared immediately
2. ___ The negative effects have persisted, but have faded with time
3. ___ The negative effects were permanent

7. In general, how would you describe the impact of the period in which your child was sick on your couples' relationship?

1	*2*	*3*	*4*	*5*	*6*	*7*
This period has distanced us / has been detrimental to our relationship			*This period had no effect on our relationship*			*This period has brought us closer / strengthened our relationship*

General Discussion

The overarching aim of the present thesis was to further our understanding of the long-term dyadic adjustment of couples whose child has been diagnosed and treated for acute lymphoblastic leukemia (ALL). In doing so, it helped expand the scope of our understanding of couples' adjustment experiences in this emergent field to two time points within the illness trajectory that have received less empirical focus: at the end of treatment (study 1) and during the survivorship period (study 2). Using a dyadic approach (Kenny et al., 2006) and based on a theoretical model of family resiliency (McCubbin et al., 2002), this research proposed a compelling approach for understanding both partners' adjustment, accounting for interdependence between partners' adjustment (Kenny et al., 2006). Thus far, only two other research teams worldwide have used this particular dyadic approach with couples in pediatric cancer settings (Compas et al., 2015; Hall, 2010a; Hall, 2010b). However, none of the aforementioned studies examined couples' relationship adjustment, making the two studies within this thesis the first empirical reports to do so. The first study aimed to determine whether both partners' reports of family functioning and parental mood near the time of their child's ALL diagnosis were related to their own and their partner's marital adjustment at the end of their child's ALL treatment. The second study was an extension of the first study. It specifically aimed to determine whether both partners' reports of relationship changes during treatment were related to their own and their partner's psychological and relationship adjustment during the survivorship period.

Interpretation of findings

Clinical distress

On a descriptive level, most parents of children with ALL in both our samples were well adjusted in that they did not meet thresholds indicative of clinical levels of psychological and relationship distress. Nevertheless, a subgroup of parents did meet these clinical cut-offs. In our first study, which was longitudinal in nature, we found that 25.5% of mothers and 21.3% of fathers reported clinical levels of marital distress at the time of their child's leukemia diagnosis, while 36.2% of mothers and 42.6% of fathers reported clinical marital distress at the end of treatment (two years later). Then in our second study, which was cross-sectional in nature, 21.4% of mothers and 20.4% of fathers reported clinical relationship distress on average 15 years following their child's leukemia diagnosis (i.e., during the survivorship period). Also, parents reporting clinical levels of psychological distress ranged from 2.9% to 13.6% for mothers and 6.8% to 9.7% for fathers. For both parents, the highest rates of psychological distress during the survivorship period were found on somatization symptoms. These findings on psychological distress during the survivorship period largely parallel those reported by both a recent review and cross-sectional study of parents of childhood cancer survivors (Ljungman et al., 2014; Malpert et al., 2015).

Together these findings seem to suggest that the couple's relationship might be quite strong at diagnosis, perhaps due to a perceived need for couples to be resilient and work together in the face of hardship. However, over time it appears that couples' relationship often gets pushed aside in an attempt to prioritize caring for the ill child. In fact, some parents qualitatively described that their relationship as a couple had changed and that although they still lived together they felt less like a couple following their child's cancer diagnosis (Björk,

Wiebe, & Hallström, 2005). Also, a recent multicenter cross-sectional study on parents of children with cancer reported that parents indicated that they felt the most emotionally connected to their partner at diagnosis and the least emotionally connected to them at the beginning and end of treatment (Wiener et al., 2016). Hence, this emotion disconnect could explain why more parents in our studies are reporting clinically significant distress at the end of their child's cancer treatments than at the time of diagnosis or during the survivorship period. It is also possible that increased caregiving demands (e.g., monitoring their child's health status) placed on parents paired with a greater distance from the healthcare team translated into greater distress for parents at the end of their child's treatments.

Relationship adjustment at the end of treatment and during survivorship

Serving as an initial test of the actor-partner interdependence model, our first study investigated longitudinal, dyadic associations between partners' mood states and family functioning at the time of their child's ALL diagnosis and both partners' marital adjustment at or nearing the end of treatment (i.e., two years post diagnosis). Altogether, findings from this study suggest that couples' long-term adjustment to pediatric leukemia is multifaceted, in that it is both an individual and interdependent experience. At the end of treatment, mothers' marital adjustment was only self-related (i.e., related to their own perception of family functioning and mood state), whereas fathers' adjustment was mostly partner-related and to a lesser extent also self-related. As we expected, better marital adjustment for mothers at the end of treatment was associated with them reporting more family support and less role conflict and role overload at diagnosis. Thus, for mothers having plenty of support from their family appears to be adaptive in fostering better marital adjustment for them up to two years later. Surprisingly, this was not the case for fathers. According to a review conducted by Clarke et

al. (2009), mothers seek more social support than fathers, thus suggesting that perhaps the significant effect that we found for family support for mothers actually reflects the fact that they were actively seeking more social support than fathers. Also, for mothers being less conflicted and overwhelmed by their various roles at diagnosis was adaptive in predicting their marital adjustment two years later. In this sense, having a clear and equitable division of caregiving and family responsibilities was very important for mothers' relationship adjustment later.

Similarly, better marital adjustment for fathers two years following their child's ALL diagnosis was related to them self-reporting less role conflict at diagnosis. Counter to our predictions, for fathers, better marital adjustment two years later was related to their own reports of greater role ambiguity and fatigue at the time of diagnosis. This could be interpreted as meaning that heightened fatigue and unclear role expectations among fathers at the time of their child's leukemia diagnosis are normal and simply reflect changing family dynamics, which often involve significant gender role adaptation for fathers (Hall, 2010a). Following their child's diagnosis, fathers often take on new childcare and household responsibilities that can require considerable time and adaptation (Clarke et al., 2009; Long & Marsland, 2011), but can be beneficial for the couple's relationship in the long-term. Indeed, lack of clarity regarding fathers' expected roles might imply that they are in fact more supportive and actively involved even early in the illness trajectory. In turn, fathers' active involvement can help reinforce their resilience as a couple and positively impact both partners' relationship satisfaction.

By finding a significant partner effect, this first thesis study empirically demonstrated that marital adjustment is an interdependent and interactional process in parents whose child

was diagnosed with leukemia. In specific, we found that greater marital distress for fathers two years post diagnosis was associated with mothers' perceiving more role conflict at diagnosis. In this way, our study seems to suggest that mothers' adjustment is exclusively self-related, whereas fathers' adjustment is both self and other-related. It is possible that mothers are expressive with their partner about feeling conflicted in their role as the primary caregiver for the ill child, and that this stress carried over into their relationship with their partner. Also, since past research has indicated that fathers depend primarily on their relationship with their partner for support (Lavee, 2005; Leventhal-Belfer, Bakker, & Russo, 1993), stresses associated with the child's illness might be translated into decreased relationship satisfaction in fathers. Interestingly, the reverse interrelationship was found in another previous longitudinal study in this field. According to this longitudinal study on 124 parents of children with cancer that was conducted by Hoekstra-Weebers et al. (1998), it was mothers' marital distress that was other-related and fathers' marital distress that was self-related. For mothers' their marital distress at one year post diagnosis was predicted by their partner's emotion-focused coping and discrepancies in emotion-focused coping styles in the couple at diagnosis, whereas fathers' marital distress one year post diagnosis was predicted by their own distress and emotion-focused coping at diagnosis. Perhaps findings from this previous longitudinal study went counter to findings from our studies due to use of different predictors for relationship adjustment and different times of assessment given that their study was during active treatment (i.e., one year post diagnosis) whereas our studies were at the end of treatment (2 years post diagnosis) and during the survivorship period (M = 15 years post diagnosis). Although, another longitudinal report of 42 couples of children with cancer found that for both partners their marital adjustment at 20 months post diagnosis was predicted in part by their

spouse's marital satisfaction at this same time (Dahlquist et al., 1996). Although the time frame of this study and our first study were relatively similar (varying only by roughly 4 months), these differences in findings can possibly be attributed to differences in the nature of our predictors. Additional empirical studies are needed in order to clarify the extent to which adjustment is self-related and partner-related.

Using a cross-sectional design and reflecting the experiences of 103 couples, our second study included in the present thesis partially replicates the interrelationship trends that were found in our first study. Once again we found that mothers' relationship adjustment was primarily self-oriented, whereas fathers' relationship adjustment was primarily other-oriented. On the one hand, greater relationship satisfaction for mothers at follow-up (i.e., during the survivorship period) was associated with them self-reporting positive relationship changes following their child's cancer treatments (i.e., improvements in intimacy, quality of partner support, sexuality, relationship satisfaction, and an overall positive impact of illness on the couple). On the other hand, fathers' reports of greater relationship satisfaction were associated with mother reporting positive relationship changes following their child's treatments (i.e., improvements in quality of partner support, conflict, relationship satisfaction, and an overall positive impact of illness on the couple). These findings suggest that although fear of cancer relapse and late effects might trouble both partners in the survivorship period, mothers' might be dealing with these concerns and adapting individually, while fathers' are turning to their partner for support and guidance in navigating these concerns and their adjustment. Given that mothers are typically the primary caregivers at the forefront of interactions with their child's healthcare team, it is possible that fathers are using mothers' views as a gauge for the illness experience and its challenges. If fathers perceive that mothers are struggling to adjust to their

new roles or appear to be unsatisfied in their relationship as a couple, then this distress can be translated to fathers and subsequently affect their psychological and relationship adjustment.

Psychological adjustment during survivorship

In our second study, we also examined how changes in relationship dynamics that were retrospectively recalled from the time of the child's treatment were associated with current psychological distress (i.e., reported during the survivorship period). Findings from this study suggest that, for both parents, recalling that the child's illness had an overall positive impact on their relationship as a couple was associated with self-reporting more psychological distress during the survivorship period (notably, more anxiety symptoms among mothers, and more depression and somatization symptoms among fathers). It is possible that these associations are reflecting state-based psychological distress, as the very nature of this follow-up study could explain parents' heightened psychological distress symptoms. When parents are anticipating their child's upcoming medical follow-up appointments, illness-concerns such as fear of relapse or late effects could greatly confound or bias the distress that they report on the day of the study. Future studies need to assess psychological distress both on a state and trait basis over time to rule out this confounding possibility in our study. One interrelationship was also reported, namely when mothers recalled that the illness had a positive impact on their time and activities with their partner, fathers tended to report more current somatization symptoms. This interrelationship is in the same direction as all the other partner effects that were reported in these two thesis studies, thus suggesting consistency in the directionality of effect and that mothers appear to be significantly influencing fathers' adjustment. Future studies will need to use longitudinal designs or interventions to assess how partners' psychological adjustment can vary across their child's illness trajectory and whether

psychological counselling early in the illness trajectory (e.g., at diagnosis or during treatment) can improve partners' adjustment later (e.g., at the end of treatment).

Impact of Cancer on the Couple: Agreement and longevity of negative effects

Although pediatric leukemia can be conceptualized as a dyadic stress for parents as a couple, it is possible that the illness might not affect both partners in the same way. To account for this possibility, we tested agreement between both partners on all predictor (relationship dimensions) and outcome (relationship satisfaction and psychological distress) variables that were used in our second study. Partners exhibited excellent agreement on the outcome variable of relationship satisfaction, and good agreement on relationship dimension predictors of sexuality, relationship satisfaction, and the overall impact of the illness on the relationship. Thereby, this suggests that mothers and fathers share a common perception of their relationship functioning and the impact of their child's illness on specific dimensions of their relationship.

Parents in our study also typically agreed on the nature of relationship changes (i.e., negative change, no change, or positive change) following their child's leukemia diagnosis. The longevity of negative relationship changes was also reported (i.e., the extent to which these negative effects endured once the child's cancer treatments ended). Negative relationship changes could be categorized as: effects that disappeared immediately following treatment, effects that faded with time, or effects that were permanent. Relationship aspects that were strengthened following the child's leukemia diagnosis included: quality of partner support, relationship satisfaction and the overall impact of the illness on the relationship. Our findings on the overall impact of the illness on the couple parallel qualitative findings from both a mixed methods study on parents of children with cancer that are at least two years post

treatment (Quin, 2005), and a qualitative study on parents' experiences when their child is five or more years post treatment or has deceased (Ljungman et al., 2016). In the qualitative report, parents specifically described that they felt that their marital relationship had been strengthened following their child's illness (Ljungman et al., 2016). Likewise, in the mixed methods report nearly three quarters of parents felt that their relationship as a couple had been strengthened by their child's illness (Quin, 2005). One partner described this in an in-depth interview as, *"I'd say it definitely grew us closer in our marriage. We just realised that we needed each other to get through it"* (Quin, 2005, p. 141). Consistent with other studies on couples' relationships in relation to pediatric cancer (Lavee, 2005; Lavee & Mey-Dan, 2003), we found that couples in our study also reported deteriorations in sexuality. Although most parents reported that negative effects on their sexuality faded with time, 8.6% of mothers and 16.9% of fathers reported that this effect was permanent. Both partners also frequently reported negative effects on intimacy and time and activities with their partner. For both these relationship aspects mothers and fathers tended to report that these negative effects either disappeared immediately following treatment or faded with time. This specification on duration of negative effects allowed us to conclude that among parents reporting that their child's illness had a negative impact on a relationship dimension, most of them reported that these negative effects faded with time. Thus, relationship difficulties appear to be reported as a transitory phase and couples eventually overcome the relationship difficulties that result from their child's illness.

Theoretical implications

From a theoretical standpoint, this thesis challenged the traditional tendency to use individual statistical approaches to understand the relationship adjustment of parents of

pediatric cancer patients. Although an integrative review on couples' functioning in the pediatric cancer context suggests that dyadic analyses should be used with quantitative measures in this field in order to understand variability between partners (da Silva et al., 2010), this analysis approach has rarely been applied in practice. For instance, in a longitudinal study on parents of children with cancer that used hierarchical multiple regression models for predicting marital adjustment at 20 months post diagnosis, these analyses were conducted separately for each partner (Dahlquist et al., 1996). Although the spouse's marital satisfaction was entered as a predictor in both models, the models themselves were not dyadic and did not account for possible interrelationships between partners' predictors and outcome variables. This approach interprets each partner's adjustment experiences are being unique and unrelated to one another. To our knowledge, only one study on couples' functioning in pediatric cancer settings has used an approach that considers the dyad as the unit of analysis and accounts for partners' interdependent perceptions. Specifically, Lavee and Mey-Dan (2003), used a repeated-measures multivariate analysis of variance (MANOVA) to determine possible gender differences in both parents' retrospective reports of change in marital quality following their child's diagnosis. In doing so, they found that husbands reported more positive change in their marital quality following their child's cancer diagnosis than wives did (Lavee & Mey-Dan, 2003). Thus, perhaps fathers are more likely to view the cancer experience as being transformational or fostering relationship growth for the couple.

Our present work further demonstrated the utility of a dyadic analysis approach for understanding couples' adjustment to the dyadic stress of pediatric leukemia. Notably, dyadic statistical approaches will likely be more sensitive to the complexity and interconnected nature of the cancer experience for couples, because they statistical account for gender differences

and the interdependent nature of couples' data (i.e., the possibility that one partner can influence the other partner; Kenny et al., 2006). This interdependence is important in parents' adjustment to pediatric cancer, because typically one parent stays at the hospital with the ill child while the other parent maintains employment and cares for the rest of the family. In this way, one parent often serves as a filter relaying cancer-related information to the other parent. Intuitively, we would expect that this communication about the child's illness is important for both parents' adjustment and that if a partner feels isolated or left out of this process, he or she might report greater marital distress or difficulties later as a result. Findings from our studies suggested that better long-term relationship adjustment for fathers was largely associated with mothers' perceptions earlier in the illness, hence the above filter hypothesis appears to be supported, and it is mothers acting as a filter in couples' experiences. Future research should examine this filter hypothesis by assessing parents' division of labour and roles following their child's diagnosis, including which parent was the primary caregiver relaying illness-related information to the other parent and the impact that these roles had on their own and their partner's adjustment later.

Using the family resiliency model (McCubbin et al., 2002), this thesis examined specific individual and relational aspects, which might underlie couples' long-term psychological and relationship adjustment to pediatric leukemia. This model included core considerations, which could affect a family's ability to be resilient when faced with a crisis event (e.g., pediatric ALL), such as their strengths and vulnerabilities (operationalized as parents' mood states and family functioning in our first study) and the meaning that they as ascribe to such an event (operationalized as the impact of the child's illness on various dimensions of the couples' relationship in our second study). This model should be further

tested in empirical studies of parents of children with cancer. Future research should specifically focus on the role of dyadic coping as a potential mediator in the association between parents' distress regarding their child's illness and their adjustment later. In addition, future research could also focus on benefit finding and posttraumatic growth as core indicators of the meaning that parents and couples have attributed to their child's illness. It would be interesting to see whether these two constructs of positive psychology are significantly relate to the partners' adjustment.

Clinical implications

Altogether, findings from our studies suggest that the long-term predictors of the psychological and relationship adjustment of mothers and fathers of children with ALL are mostly unique to each parent, with the exception of a few select predictors that are shared by both parents. At the end of the child's treatment, couples face challenges related to adjusting to their life post cancer. Children have less frequent medical follow-up and parents have more responsibility in monitoring their child's health (Muskat et al., 2017). Actually, couples of children with cancer report feeling most emotionally connected to their partner at diagnosis and least emotionally connected to them at the beginning and end of treatment (Wiener et al., 2016). Indeed, a parent in a recent qualitative study described this as feeling less like a couple and more like "*two fighters side by side*" (Martin et al., 2016). Clinically speaking, this seems to suggest that although most couples feel very connected to one another, when faced with the initial crisis (i.e., diagnosis), they may lose this connection and grow apart over time. This proposition is supported by the fact that in our sample clinical levels of marital distress were met by 25.5% of mothers and 21.3% of fathers at diagnosis, but 36.2% of mothers and 42.6% of fathers at 2 years post diagnosis. According the normative sample used in the construction

and validation of the Marital Adjustment Test (which was also used in our study), 48 out of 236 participants (118 husbands and 118 wives all of which were married and most which had one child or were childless) were considered to be maladjusted, with 17% of them meeting the cut-off score indicative of clinical marital distress (Locke & Wallace, 1959). Being able to conserve the emotional connection between partners throughout the illness trajectory should help reduce their marital distress later. Also, to foster better adjustment for couples later on, the family unit should provide ample support for both parents' (particularly mothers) at diagnosis. Shortly following diagnosis, the child's health care team could also provide brief, educational sessions to help parents learn how to balance their pre-existing parental responsibilities with their new caregiving ones. In turn, this could assist couples in having fewer role-related difficulties (e.g., less role conflict) and stronger marital adjustment later on. Open discussions between partners might also allow for a more equitable division of parenting and caregiving responsibilities, and reduced relationship strain in the long run. Given their support needs and the rearrangement of their parental roles, couples would also likely benefit from couples-based interventions. Couples' counselling sessions should be provided regularly during all illness phases (i.e., at diagnosis, during active treatment, at the end of treatment, and during the survivorship period). Interventions to help couples reconnect with one another emotionally, and providing them with avenues to discuss their relationship and its associated strengths and difficulties are expected to be most beneficial. Accordingly, past research on couples coping with one partner's chronic illness have studied such interpersonal strategies and referred to them as 'relationship talk' or 'social sharing' (Badr & Acitelli, 2005; Boinon et al., 2014). By encouraging partners to openly discuss their relationship and express their thoughts and concerns regarding their child's illness, partners could identify their sources of

strength and interpersonal difficulties early on. It is also important that clinicians' explain to couples that the stresses related to their child's illness might present themselves in different ways for each partner, thus implying that their needs might vary somewhat. Partners should be encouraged to be mindful of this and respect differences in how themselves and their partner are adjusting to this hardship. Also, some parents might benefit from individual counselling, so that each partner's respective needs and concerns can be addressed and areas of individual difficulty can be worked on.

As indicated in our second study, partners' recalling positive relationship changes as having occurred during their child's treatments were associated with them reporting better relationship adjustment during the survivorship period. For instance, mothers' recalling more positive change on the quality of support that they received from their partner was related to them self-reporting greater relationship satisfaction on average 15 years after the end of their child's treatment. For this reason, it is essential for couples' therapy clinicians to clearly identify both the aspects that have improved and deteriorated following the child's cancer diagnosis. Hence, clinicians will be able to tailor their therapy sessions to work on the specific relationship aspects that the couple is struggling with. On the contrary, findings from our study also demonstrated that feeling that the child's illness was associated with a strengthened relationship as a couple might not be associated with a favourable outcome for either partner's psychological distress in the survivorship period. For instance, mothers' recalling that overall the child's illness had a positive impact on their relationship with their partner was associated with fathers reporting more depression symptoms during the survivorship period. Clinicians should openly discuss these types of possibilities with couples. They should explain that following a crisis event, disruptions and difficulties in individual or dyadic functioning are to

be expected and reassure them that this is not maladaptive. This clarification is important, so that parents do not feel guilty if they are overwhelmed by the demands of their child's illness. Similarly, it is important that couples feel comfortable addressing their concerns and struggles with both their partner and their clinician. Open communication and mutual support among partners is critical to foster better relationship adjustment later on.

Contributions

The present thesis addressed a few core limitations in the field of couples' relationships in relation to pediatric leukemia. First, the studies included both partners within the couple, as opposed to just focusing on the experiences of one parent. Traditionally, researchers have compartmentalized relationship changes into positive and negative effects (Lavee & Mey-Dan, 2003), and examined mothers and fathers' adjustment separately (Dahlquist et al., 1996). Also, compartmentalizing relationship changes into being either positive or negative might not be the most representative approach as it is very likely that couples' will report both positive and negative changes throughout their child's illness trajectory and that the nature of changes might depend on illness particularities. For example, parents will probably report more negative relationship changes if their child's cancer treatments have several complications versus their child's treatment is going well. It is also possible that the nature of relationship changes that parents report varies according to the stage of the illness, with parents reporting more negative changes when less time has passed since their child's leukemia diagnosis and more positive changes when significantly more time has passed. Second, our two studies are the first empirical reports to use the actor-partner interdependence model to understand couples' relationship adjustment as no previous dyadic studies in this field have specifically examined relationship adjustment. The most marked contributions of this thesis are our

findings, which clearly demonstrate the interactional and interrelated nature of couples' adjustment to pediatric cancer. In fact, these interrelations show that in order to truly understand couples' adjustment we should consider both partners' perspectives and experiences simultaneously. Failure to do so would result in an incomplete portrayal of how couples adjust to the dyadic stress of their child's illness.

Finally, we reported the first empirical data on the *Impact of Cancer on the Couple*. This self-report questionnaire was specifically designed for use with this cohort of couples with children diagnosed and treated for ALL. This measure provided a retrospective report of the impact of the child's cancer on several core relationship dimensions. To date, only two other quantitative reports (both conducted by the same research team) have used a similar adapted measure to retrospectively assess marital quality and the nature of changes that partner perceived following their child's cancer diagnosis (Lavee, 2005; Lavee & Mey-Dan, 2003). Although this measure shared a few relationship dimensions such as sexuality and conflict resolution with the measure used by the previous research team, there were a few fundamental differences. First, this questionnaire was constructed following a targeted reflection on couples' relationship functioning following their child's cancer diagnosis and what relationships aspects past researchers typically report as being most affected by pediatric cancer (da Silva et al., 2010; Long & Marsland, 2011). In doing so, this questionnaire examined relationship aspects that were not assessed by these two previous reports, such as quality of partner support, intimacy, and overall relationship satisfaction. Data provided from this new questionnaire also contributed to the advancement of this field by its precision regarding the duration of couples' negative relationship changes following their child's ALL diagnosis. By knowing whether these negative changes disappeared immediately following the

end of the child's treatments, faded with time, or were permanent, we are better able to understand the temporal process behind couples' adjustment to pediatric cancer. Future studies should use this questionnaire to assess temporal changes in couples' relationship dynamics at critical stages within the illness trajectory: at diagnosis, during treatment, at the end of treatment, and in the survivorship period. Doing so, would allow us to identify when couples experience the most relationship difficulties and which particular aspects have the most enduring negative effects on couples. It would also be helpful to administer this questionnaire to parents of children with more sombre cancer prognoses (e.g., parents of children with brain tumours) to see if their relationship functioning differs or resembles that of parent of children with ALL.

Limitations and future directions

Both respective studies have select methodological limitations. To begin with, the research program of the present thesis examined the two pathways of the *Resiliency Model of Family Stress, Adjustment and Adaptation* (McCubbin et al., 2002) separately. One could consider testing both pathways in this model simultaneously using a structural equation modelling approach. In this way, we would be able to see the relative contributions of couples' resources and the meaning that they have given to the illness in explaining their overall adjustment.

We should be also cautious about overextending our findings, especially given that our dyadic analyses were exploratory in nature and both our samples are homogenous in that they exclusively assessed the experiences of parents of children with ALL who stayed together. Our samples reflect only one diagnosis category of childhood cancers, which has a relatively good overall prognosis (5-year survival >80%) and a standardized treatment regime lasting

approximately two years. Consequently, it is possible that our findings would not translate to parents of children with more sombre cancer prognoses (e.g., parents of children with brain tumours). Due the increased clinical severity of these children's cancer, we can expect that these parents would have reported significantly greater psychological and marital distress and more parents would have met cut-offs for clinical levels of distress than the parents in our samples. In both our studies, we excluded parents of children with ALL that had experienced a second cancer or deceased, since we expected that their experiences were too distinct to justify confounding them together with parents of children with better treatment outcomes. Also, both our samples were Quebec cohorts of predominately French-speaking parents, with the vast majority being well adjusted and only a small subgroup reporting clinical levels of psychological and relationship distress. This significantly limits the generalizability of our findings. In this sense, our samples lacked cultural diversity, which could have significantly impacted parents' division of caregiving and family responsibilities, as well as the ways couples related to each other. It is possible that in another culture, the interrelationships that we found would have been different or more nuanced. For example, we found that fathers' long-term adjustment was significantly related to mothers' predictors earlier in the illness trajectory (e.g., at diagnosis). However, it is possible that in another culture, it could have been mothers' adjustment that was related to fathers' predictors. Perhaps the significant interrelationships that we found were not reflecting gender roles, but instead were reflecting who was the primary caregiver translating the illness details and experience to the other partner. Future research should explicitly examine the division of caregiving and family responsibilities and its contribution to explaining each partner's adjustment. Lastly, although all eligible couples were contacted for recruitment, our results only reflect those who

participated. It is possible that couples that were experiencing heightened distress or significant difficulties at the time of the study declined participating. However, given the demands associated with having a chronically ill child, the attrition rates that are expected for follow-up studies, and the necessity of both partners participation, the final participation rates in both our studies were quite respectable (61% and 46%, respectively).

It is also important to be mindful of our study design limitations. In our first study, we led a secondary analysis of longitudinal data from a Quebec cohort of children with ALL and their parents. Thus, we needed to work within the constraints of the variables and data that were already collected. Given our underlying theoretical model, we selected the three constructs (parental mood states, family functioning, marital adjustment) that we felt would best encapsulate the components of this theoretical model in order to describe parents' experiences of having their child diagnosed and treated for ALL. Also, since this study was an exploratory secondary analysis and our APIM models required data from both partners at both time points (diagnosis and 2 years post diagnosis), our sample was reduced from an initial recruitment of 138 families to 47 couples meeting all eligibility criteria. This meant that we had reduced statistical power, which might have prevented us from finding other meaningful results that would have been significant with a larger sample. Even so, this study served as an appropriate preliminary test of the interdependence model for understanding couples' dyadic adjustment in relation to pediatric cancer. Next, we should be aware that in our second study each parent retrospectively reported the relationship changes that recalled during their child's cancer treatments. Also, given the cross-sectional nature of this study, causal associations and the directionality of effects could not be determined. In this way, it is viable that parents' current adjustment (both in terms of psychological distress and relationship satisfaction)

biased their recall of past events relating to their child's illness (Blome & Augustin, 2015; Wilson & Ross, 2003). To address this possibility, future research should examine associations between cancer-related relationship effects and adjustment using a longitudinal design instead.

Taking into account the above-mentioned limitations and the current state of the literature on couples' functioning in relation to pediatric cancer, a few core avenues should be taken by future research. First, future research should prioritize using a dyadic, longitudinal design to further investigate the process issues involved in couples' adjustment over their child's illness trajectory. In particular, future research should examine whether couples' common coping efforts (i.e., dyadic coping) during treatment can at least partially mitigate or explain the association between partners' perceived caregiving strain during treatment and their adjustment following the end of their child's cancer treatments. Such research should also clarify what caregiving aspects are deemed to be most demanding by each partner and what forms of support would be most appreciated. By knowing what caregiving aspects each partner struggles with the most, clinical interventions can be tailored to address each partner's unique needs, as well as the shared needs of the couple.

Conclusion

The present thesis proposed to address a few core limitations in the literature on couples' functioning in relation to pediatric cancer by purposely applying a dyadic approach to understand the dyadic, psychological and relationship adjustment of couples whose child has been diagnosed with acute lymphoblastic leukemia (ALL) at the end of treatment (Study 1) and during the survivorship period (Study 2). Our first dyadic study demonstrated the importance of both individual strengths (i.e., mood states) and the social environment surrounding the child (i.e., family functioning) at the time of diagnosis and the extent to which these longitudinal aspects are associated with stronger marital adjustment up to two years later. Our second study revealed that during the survivorship period a small subgroup of couples still report clinically significant distress, and that retrospective reports of perceived relationship changes during treatment are significantly related to couples' long-term adjustment. Together, findings from these two studies suggest that the adjustment of mothers of children with ALL is an individual experience, while the adjustment of fathers is mostly an interdependent experience. This implies that fathers' adjustment is primarily related to their partner's well-being and interpersonal experiences. Further research needs to be conducted to determine the directionality of these effects, and whether these associations are rooted in traditional or non-traditional gender-role expectations. Using a dyadic approach, this research challenges how we have traditionally conceptualized couples' adjustment to pediatric cancer. Instead it provides a strong, empirical basis for interpreting couples' adjustment to pediatric leukemia as a dyadic stress, influencing both partners in distinct and interdependent ways.

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Appendix A. Ethical approvals for QcALL 91-95 database of ALL study at Sainte-Justine UHC and PETALE study.

Le 16 mars 2011

Monsieur Philippe Robaey
Direction de la recherche
CHU Sainte-Justine

Objet : Acceptation de votre projet de recherche

CHU Sainte-Justine
Le centre hospitalier universitaire mère-enfant

Pour l'avenir des enfants

Université de Montréal

Monsieur,

C'est avec plaisir que nous acceptons votre demande de révision de dossiers médicaux dans le cadre de votre projet de recherche intitulé « Patients soignés au CHU Sainte-Justine pour une leucémie lymphoblastique aiguë. ». Cette étude se fera pour les dossiers ouverts entre le 1er janvier 1993 et le 31 décembre 1999. Cette recherche se fera en collaboration avec Mme Sophie Marcoux, Dr Maja Krajinovic et Dr Caroline Laverdière.

Par la présente, je vous autorise à prendre connaissance des dossiers à des fins d'étude ou d'enseignement, sans le consentement du titulaire de l'autorité parentale du patient concerné, en vertu de l'article 19.2 de la Loi dans la mesure où les conditions suivantes sont respectées.

Je comprends que votre projet respecte les normes d'éthique ou d'intégrité scientifique généralement reconnues en matière d'étude ou d'enseignement et que vous respecterez le caractère confidentiel des renseignements que vous obtiendrez. De plus, nous vous demandons de prendre les moyens requis pour qu'aucun renseignement permettant d'identifier le patient ne soit utilisé ou communiqué lors de cette recherche.

Cette autorisation est valable jusqu'au 16 mars 2012 et ne vaut que pour les fins qui y sont énoncées.

Parallèlement, si vous deviez adresser votre projet au comité d'éthique à la recherche, nous vous recommandons d'y joindre cette lettre.

Par ailleurs, nous transmettons une copie de cette acceptation à la coordonnatrice des archives médicales, qui vous aidera dans votre démarche. Si plusieurs dossiers sont archivés à l'extérieur, elle vous en communiquera les frais.

Nous vous prions d'agréer, Monsieur, l'expression de nos sentiments les meilleurs.

Le Directeur des affaires médicales et universitaires,

[redacted]

Marc Girard, M.D.

MG/md

c. c. Coordonnatrice des archives médicales
M. Jean-Marie Therrien, président au comité d'éthique à la recherche

p.s. Veuillez vous présenter aux archives médicales avec cette lettre.

Tél.: 514 345-4625 / Téléc.: 514 345-4805

3175, Côte-Sainte-Catherine
Montréal (Québec)
H3T 1C5

Le 04 janvier 2013

Docteure Caroline Laverdière
Hémato/Onco
Étage 2e Bloc 6

OBJET: Titre du projet: PETALE Prévenir les Effets tardifs des Traitements de la ALL chez l'Enfant

No. de dossier: 3607

Responsables du projet: Caroline Laverdière M.D., chercheurs responsable. Chercheurs principaux: Daniel Sinnett et Maja Krajinovic. Collaborateurs: Nathalie Alos, Gregor Andelfinger, Emile Levy, Sarah Lippé, Philippe Robaey et Serge Sultan

CHU Sainte-Justine
Le centre hospitalier universitaire mère-enfant

Pour l'amour des enfants

Université de Montréal

Chère Docteure,

Votre projet cité en rubrique a été approuvé par le comité d'éthique de la recherche en date du 21 décembre 2012. Vous trouverez ci-joint la liste des documents approuvés ainsi que votre formulaire d'information et de consentement estampillé dont nous vous prions de vous servir d'une copie pour distribution. Notez que pour une collaboration avec un (ou plusieurs) tiers (institutions ou entreprises privées) impliquant des transferts de fonds et/ou données et/ou matériel biologique, une entente (contrat) doit être conclue avec le Bureau des ententes de recherche (BER).

Tous les projets de recherche impliquant des sujets humains doivent être réexaminés annuellement et la durée de l'approbation de votre projet sera effective jusqu'au **21 décembre 2013**. Notez qu'il est de votre responsabilité de soumettre une demande au comité pour que votre projet soit renouvelé avant la date d'expiration mentionnée. Il est également de votre responsabilité d'aviser le comité dans les plus brefs délais de toute modification au projet ainsi que de tout effet secondaire survenu dans le cadre de la présente étude.

Nous vous souhaitons bonne chance dans la réalisation de votre projet et vous prions de recevoir nos meilleures salutations.

[redacted]

Jean-Marie Therrien, Ph.D., éthicien
Président du Comité d'éthique de la recherche

JMT/mhl
c.c. BER

3175, Côte-Sainte-Catherine
Montréal (Québec)
H3T 1C5

Appendix B. Measures and Questionnaires

Study 1:

Locke-Wallace Marital Adjustment Test (MAT) (Locke & Wallace, 1959)

Profile of Mood States-Bipolar Form (POMS-BI) (Lorr, McNair, & Heuchert, 2003)

Family Well-Being Assessment (FWA) (Caldwell, 1988)

Study 2:

Sociodemographic questionnaire

Brief Symptom Inventory-18 (BSI-18) (Derogatis, 2000)

Impact of Cancer on the Couple (Péloquin & Sultan, 2013)

Dyadic Adjustment Scale (DAS-4) (Sabourin, Valois, & Lussier, 2005)

QUESTIONNAIRE SUR LA VIE EN COMMUN

Code:_____ Date d'aujourd'hui: _____

#Dossier: _____ Mère ou père:_____

Veuillez cocher, sur l'échelle ci-dessous, le point qui décrit le mieux le degré de bonheur qui existe dans votre mariage actuel (ou dans votre relation si en cohabitation). Le point central, "heureux" représente le degré de bonheur que la plupart des gens éprouvent au cours de leur union maritale (ou relation). L'échelle s'étend graduellement d'une part vers le petit nombre de personnes dont le mariage (la relation) est très malheureux et d'autre part, vers le petit nombre qui vivent ensemble dans un bonheur absolu.
S.V.P encercler votre réponse ici.

• • • • • • •

Très malheureux Heureux Parfaitement heureux

Veuillez indiquer, pour chaque point suivant, le degré approximatif d'accord ou de désaccord entre vous et votre partenaire, depuis les 12 derniers mois. Cochez la colonne qui correspond le mieux à votre opinion.

	Toujours d'accord	Presque toujours d'accord	Désaccord occasionnel	Désaccord fréquent	Presque toujours en désaccord	Toujours en désaccord
2. Administration du budget familial						
3. Loisirs						
4. Témoignages d'affection						
5. Ami(e)s						
6. Relations sexuelles						
7. Conventions sociales (conformité aux exigences de la société)						
8. Philosophie de la vie						
9. Relations avec la belle-famille						

10. Quand il y a désaccord entre vous :
_____ C'est l'homme qui cède
_____ C'est la femme qui cède
_____ Vous arrivez à établir un compromis

11. Est-ce que vous et votre partenaire avez des activités ensemble dehors de la maison?
_____ toutes
_____ quelques unes
_____ très peu
_____ aucune

12. Pour occuper vos moments de loisirs que préférez-vous généralement?
_____ des activités extérieures
_____ rester à la maison

Votre partenaire préféré-t-il/elle?
_____ des activités extérieures
_____ rester à la maison

13. Avez- vous déjà souhaite ne pas être mariée (ou être en cohabitation)?
_____ fréquemment
_____ occasionnellement
_____ rarement
_____ jamais

14. Si vous pouviez recommencer votre vie, pensez-vous que vous voudriez :
_____ épouser (ou être avec) la même personne
_____ épouser (ou être avec) une autre personne
_____ ne pas vous marier (ou vivre ensemble)

15. Vous confiez-vous à votre partenaire :
_____ presque jamais
_____ rarement
_____ très souvent
_____ toujours

ÉCHELLE D'HUMEURS

Code:_____ Date d'aujourd'hui: _____

#Dossier: _____ Mère ou père:_____

Ci-dessous se trouve une liste de mots décrivant différentes façons dont les gens peuvent se sentir. Lisez chaque mot attentivement. Veuillez indiquer le dégrée auquel vous avez ressenti chaque émotion dans les derniers SEPT JOURS en entourant le chiffre qui correspond le mieux à votre expérience.

La signification de chaque chiffre est la suivante:
0 = Je ne me sentais PAS comme ça
1 = Je me sentais TRÈS PEU comme ça
2 = Je me sentais PLUTÔT comme ça
3 = Je me sentais BEAUCOUP comme ça

1.	Tranquille	0	1	2	3	29.	Agité(e)	0	1	2	3
2.	En colère	0	1	2	3	30.	Désorienté(e)	0	1	2	3
3.	Enjoué(e)	0	1	2	3	31.	Énergique	0	1	2	3
4.	Faible	0	1	2	3	32.	Seul(e)	0	1	2	3
5.	Tendu(e)	0	1	2	3	33.	Compatissant(e)	0	1	2	3
6.	Confus(e)	0	1	2	3	34.	Exténué(e)	0	1	2	3
7.	Gai(e)	0	1	2	3	35.	Puissant(e)	0	1	2	3
8.	Triste	0	1	2	3	36.	Attentif(ve)	0	1	2	3
9.	Amical(e)	0	1	2	3	37.	Serein(e)	0	1	2	3
10.	Fatigué(e)	0	1	2	3	38.	De mauvaise humeur	0	1	2	3
11.	Fort(e)	0	1	2	3	39.	Joyeux(se)	0	1	2	3
12.	Lucide	0	1	2	3	40.	Insécure	0	1	2	3
13.	Plein(e) de quiétude	0	1	2	3	41.	Fragile	0	1	2	3
14.	Maussade	0	1	2	3	42.	Perplexe	0	1	2	3
15.	Vif(ve)	0	1	2	3	43.	Actif(ve)	0	1	2	3
16.	Timide	0	1	2	3	44.	Déprimé(e)	0	1	2	3
17.	Nerveux(se)	0	1	2	3	45.	Accomodant(e)	0	1	2	3
18.	Mélangé(e)	0	1	2	3	46.	Apathique	0	1	2	3
19.	Vigoureux(se)	0	1	2	3	47.	Fonceur(se)	0	1	2	3
20.	Abattu(e)	0	1	2	3	48.	Capable de me concentrer	0	1	2	3
21.	Gentil(le)	0	1	2	3	49.	Calme	0	1	2	3
22.	Épuisé(e)	0	1	2	3	50.	Fâché(e)	0	1	2	3
23.	Audacieux(se)	0	1	2	3	51.	Réjoui(e)	0	1	2	3
24.	Compétent(e)	0	1	2	3	52.	Incertain(e)	0	1	2	3
25.	Paisible	0	1	2	3	53.	Anxieux(se)	0	1	2	3
26.	Furieux(se)	0	1	2	3	54.	Mêlé(e)	0	1	2	3

27.	Coeur léger	0	1	2	3	55.	Plein(e) d'entrain	0	1	2	3
28.	Indécis(e)	0	1	2	3	56.	Découragé(e)	0	1	2	3
57.	Bonne nature	0	1	2	3	65.	Mal à l'aise	0	1	2	3
58.	Las(se)	0	1	2	3	66.	Stupéfait(e)	0	1	2	3
59.	Confiant(e)	0	1	2	3	67.	Dynamique	0	1	2	3
60.	Efficace	0	1	2	3	68.	Morose	0	1	2	3
61.	Détendu(e)	0	1	2	3	69.	Affectueux(se)	0	1	2	3
62.	Irrité(e)	0	1	2	3	70.	Somnolent(e)	0	1	2	3
63.	Exalté(e)	0	1	2	3	71.	Sûr(e) de moi	0	1	2	3
64.	Inadéquat(e)	0	1	2	3	72.	Alerte	0	1	2	3

Temps de mesure: _____
Code du sujet: _____
de dossier : _____

Répondant (père ou mère) : _____
Date d'aujourd'hui : _____

Évaluation du bien-être familial
Version parents

La façon dont les membres d'une famille travaillent ensemble et s'appuient mutuellement comprend plusieurs éléments qui ne sont pas tous bien compris par les chercheurs. Les questions suivantes portent sur ce que c'est d'être un membre de votre famille. Lorsque vous remplissez ce questionnaire, soyez le plus franc possible, mais ne vous attardez pas trop sur chaque question. Les questions portent sur *votre rôle en tant que parent*. *Toutes vos réponses sont confidentielles*.

Ce questionnaire consiste en deux sections. Les catégories de réponses diffèrent quelque peu d'une section à l'autre. Avant de commencer une section, veuillez lire attentivement les catégories de réponses de la section et indiquer votre réponse en *encerclant* la réponse qui correspond le mieux à ce que vous ressentez depuis les 12 derniers mois.

Remarque : Le masculin est utilisé ici comme générique pour désigner à la fois les hommes et les femmes.

Les choix suivants s'appliquent à toutes les questions de la première section. Veuillez encercler la réponse qui décrit le mieux dans quelle mesure vous êtes d'accord ou en désaccord avec chaque énoncé.

OUI	=	FORTEMENT d'accord
Oui	=	Moyennement d'accord
oui	=	quelque peu d'accord
non	=	quelque peu en désaccord
Non	=	Moyennement en désaccord
NON	=	FORTEMENT en désaccord

1.	Je ne peux parfois accomplir mon travail sans faire des choses avec lesquelles mon conjoint serait en désaccord	**OUI**	Oui	oui	non	Non	**NON**
2.	D'un jour à l'autre, je sais ce que ma famille attend de moi en tant que parent	**OUI**	Oui	oui	non	Non	**NON**
3.	La plupart du temps, les autres membres de la famille s'attendent à ce que je sois un meilleur parent	**OUI**	Oui	oui	non	Non	**NON**
4.	Ma famille prend régulièrement le temps de discuter de sujets qui touchent la famille	**OUI**	Oui	oui	non	Non	**NON**
5.	Les membres de notre famille sont sous beaucoup de tension	**OUI**	Oui	oui	non	Non	**NON**
6.	En général, ma famille est le genre auquel je veux appartenir	**OUI**	Oui	oui	non	Non	**NON**
7.	Présentement, ma vie m'apporte beaucoup de satisfaction	**OUI**	Oui	oui	non	Non	**NON**
8.	J'ai de la difficulté à satisfaire aux demandes discordantes des membres de ma famille	**OUI**	Oui	oui	non	Non	**NON**
9.	Je sais exactement ce que ma famille attend de moi	**OUI**	Oui	oui	non	Non	**NON**
10.	Si mon conjoint est malade ou absent, ma famille a de la difficulté à s'y adapter et à pourvoir aux besoins de ma famille	**OUI**	Oui	oui	non	Non	**NON**
11.	Ma famille s'attend à ce que je fasse plus de choses à la maison que ce dont j'en suis capable	**OUI**	Oui	oui	non	Non	**NON**
12.	J'exerce une influence sur ce qui se passe au sein de ma famille	**OUI**	Oui	oui	non	Non	**NON**
13.	Je prends la plupart des décisions touchant la famille sans consulter mon conjoint	**OUI**	Oui	oui	non	Non	**NON**
14.	Mon conjoint comprend que j'ai besoin de passer du temps seul avec mes amis	**OUI**	Oui	oui	non	Non	**NON**

15.	Je trouve que ma vie est pleine d'espoir présentement	**OUI**	Oui	oui	non	Non	**NON**
16.	Je dirais sans hésitation que ma famille vit dans un climat stressant à la maison	**OUI**	Oui	oui	non	Non	**NON**
17.	Je suis extrêmement satisfait de mon rôle en tant que parent	**OUI**	Oui	oui	non	Non	**NON**
18.	Je trouve que je vis une vie très solitaire en ce moment	**OUI**	Oui	oui	non	Non	**NON**
19.	Mon conjoint et moi ne nous entendons pas sur la façon dont on devrait faire les choses	**OUI**	Oui	oui	non	Non	**NON**
20.	Faute d'argent et de temps, je ne peux faire plusieurs choses pour ma famille, bien que je le voudrais	**OUI**	Oui	oui	non	Non	**NON**
21.	On me donne assez d'information pour bien accomplir mes tâches en tant que parent	**OUI**	Oui	oui	non	Non	**NON**
22.	Même lorsque nous ne sommes pas ensemble, je sens que j'ai l'appui des membres de ma famille	**OUI**	Oui	oui	non	Non	**NON**
26.	Ma vie est plutôt vide présentement	**OUI**	Oui	oui	non	Non	**NON**
27.	Être parent me donne un sentiment de satisfaction profonde comparativement aux autres choses qui m'intéressent	**OUI**	Oui	oui	non	Non	**NON**
30.	Les autres membres de ma famille me trouvent compétent en tant que parent	**OUI**	Oui	oui	non	Non	**NON**
31.	Je pose des questions aux autres membres de ma famille et je fais souvent ce qu'ils suggèrent	**OUI**	Oui	oui	non	Non	**NON**
32.	Je ne sais pas exactement ce que ma famille pense de moi	**OUI**	Oui	oui	non	Non	**NON**
33.	Je dirais que chez nous il y a un climat de tension explosive	**OUI**	Oui	oui	non	Non	**NON**
35.	La plupart de mes amis sont des amis de ma famille	**OUI**	Oui	oui	non	Non	**NON**

36.	Ma famille fait rarement des choses ensemble pour s'amuser	**OUI**	Oui	oui	non	Non	**NON**
37.	Il m'est possible de paraître détenu et de ne pas montrer aux membres de ma famille que je me sens nerveux	**OUI**	Oui	oui	non	Non	**NON**
38.	J'écoute attentivement les autres membres de ma famille pour qu'ils sachent que je les écoute	**OUI**	Oui	oui	non	Non	**NON**
39.	J'ai trop de responsabilités à la maison	**OUI**	Oui	oui	non	Non	**NON**
40.	Je n'ai pas été adéquatement préparé à assumer les tâches quotidiennes qu'un parent a à accomplir	**OUI**	Oui	oui	non	Non	**NON**
41.	Ma vie est présentement très agréable	**OUI**	Oui	oui	non	Non	**NON**
42.	Je trouve que ça ne vaut pas la peine pour moi de faire des suggestions concernant des sujets qui touchent la famille, parce qu'on prend des décisions sans tenir compte de mon opinion	**OUI**	Oui	oui	non	Non	**NON**
43.	Si un(e) de mes bons(bonnes) amis(ies) m'annonçait qu'il(elle) est intéressé(e) à commencer sa propre famille, je serais porté, à cause de mon expérience personnelle, à lui exprimer des réserves sérieuses à ce sujet	**OUI**	Oui	oui	non	Non	**NON**
44.	Je m'adapte rapidement aux changements qui surviennent dans ma famille	**OUI**	Oui	oui	non	Non	**NON**
45.	Je trouve qu'être parent empiète sur les autres rôles que j'ai à assumer dans la vie	**OUI**	Oui	oui	non	Non	**NON**
46.	Dans ma famille, la discipline des enfants est prise en charge par un seul parent/adulte	**OUI**	Oui	oui	non	Non	**NON**
47.	Je trouve que j'ai du travail supplémentaire à la maison, au-delà de ce qui est raisonnable de s'attendre de moi	**OUI**	Oui	oui	non	Non	**NON**
48.	Mon conjoint et moi, nous nous sentons rarement frustrés dans notre rôle en tant que parents	**OUI**	Oui	oui	non	Non	**NON**

49.	Je suis prêt à faire face à toute situation qui pourrait survenir au sein de ma famille	**OUI**	Oui	oui	non	Non	**NON**
50.	Les différents membres de ma famille me demandent des choses incompatibles	**OUI**	Oui	oui	non	Non	**NON**
52.	Si on me le demandait, je pourrais définir exactement ce en quoi consiste mon rôle en tant que parent	**OUI**	Oui	oui	non	Non	**NON**

Dans la section qui suit, vous trouverez des questions sur vos réactions, ainsi que celles des autres membres de votre famille, face à des situations familiales. Pour chaque question, veuillez encercler la réponse qui décrit le mieux ce qui se passe dans votre famille.

OUI	=	Presque toujours
Oui	=	Très souvent
oui	=	Souvent
non	=	À l'occasion
Non	=	Pas très souvent
NON	=	Presque jamais

54.	J'ai des troubles d'estomac	**OUI**	Oui	oui	non	Non	**NON**
55.	Mon conjoint m'appuie et se prononce en ma faveur de mes décisions devant les autres membres de la famille et nos amis	**OUI**	Oui	oui	non	Non	**NON**
56.	J'ai de la difficulté à m'endormir ou à rester endormi	**OUI**	Oui	oui	non	Non	**NON**
57.	Ma famille porte attention à ce que je dis	**OUI**	Oui	oui	non	Non	**NON**
59.	Je suis troublé par des maux de tête	**OUI**	Oui	oui	non	Non	**NON**
60.	Je me fais beaucoup de souci pour ma famille	**OUI**	Oui	oui	non	Non	**NON**
62.	Les membres de ma famille se défendent mutuellement face aux personnes qui ne sont pas de la famille	**OUI**	Oui	oui	non	Non	**NON**

63.	Je suis troublée par des sentiments de nervosité ou de tension	**OUI**	Oui	oui	non	Non	**NON**
65.	Dernièrement, j'ai pris ou perdu du poids	**OUI**	Oui	oui	non	Non	**NON**
66.	Quand je ressens vraiment le besoin de parler à quelqu'un, les enfants de ma famille sont prêts à m'écouter	**OUI**	Oui	oui	non	Non	**NON**
67.	Mon conjoint fait attention à ce que je dis	**OUI**	Oui	oui	non	Non	**NON**
68.	Quand j'ai besoin de parler à mon conjoint, il est prêt à m'écouter	**OUI**	Oui	oui	non	Non	**NON**
69.	Si je décidais de faire les choses contrairement à la coutume de ma famille, cela créerait beaucoup de tension au sein de la famille	**OUI**	Oui	oui	non	Non	**NON**
70.	Le partage des responsabilités domestiques est une source de conflit pour mon conjoint et moi	**OUI**	Oui	oui	non	Non	**NON**
71.	Ma famille me demande mon avis sur des sujets importants	**OUI**	Oui	oui	non	Non	**NON**
72.	Je ne me sens pas prêt à assumer les tâches d'un parent	**OUI**	Oui	oui	non	Non	**NON**
73.	On m'informe des choses importantes qui touchent ma famille	**OUI**	Oui	oui	non	Non	**NON**
74.	Je ne sais pas exactement quelles sont toutes les responsabilités que j'ai à assumer en tant que parent	**OUI**	Oui	oui	non	Non	**NON**

QUESTIONNAIRE SOCIODÉMOGRAPHIQUE

1. Êtes-vous l'un des parents qui s'est occupé de l'enfant au moment du diagnostic et des traitements?

☐Oui (1) ☐Non (0)

2. Sexe : ☐Masculin (1) ☐Féminin (0)

3. Date de naissance : _____ /_____ /_____
Jour Mois Année

4. Plus haut degré de scolarité complété :

___ Secondaire non complété (1)
___ Secondaire (2)
___ Collégial (3)
___ Baccalauréat (4)
___ Maîtrise (5)
___ Doctorat (6)
___ Post-doctorat (7)
___ Autre (99), spécifiez : _____

5. Occupation principale présentement :

___ Travail à temps plein (1)
___ Travail à temps partiel (2)
___ À la retraite (3)
___ Chômage / recherche d'emploi (4)
___ À la maison (5)
___ Autre (99), spécifiez : _____

6. Quel est votre revenu personnel annuel avant déduction d'impôts?

___ Moins de 15 000$ (1)
___ 15 000 à 29 999$ (2)
___ 30 000 à 49 999$ (3)
___ 50 000 à 69 999$ (4)
___ 70 000 à 89 999$ (5)
___ 90 000 à 109 000$ (6)
___ 110 000 à 149 999$ (7)
___ 150 000$ et plus (8)

7. Quel est le revenu personnel annuel de votre conjoint avant déduction d'impôts?

___ Moins de 15 000$ (1)
___ 15 000 à 29 999$ (2)
___ 30 000 à 49 999$ (3)
___ 50 000 à 69 999$ (4)
___ 70 000 à 89 999$ (5)
___ 90 000 à 109 000$ (6)
___ 110 000 à 149 999$ (7)
___ 150 000$ et plus (8)

8. Langue maternelle :

___ Français (1) ___ Anglais (2) ___ Autre (99), spécifiez : _____

9. Pays de naissance :
___ Canada (1) ___ Autre (99), spécifiez : _____

10. Si vous êtes née à l'extérieur du Canada, depuis combien d'années vivez-vous au Canada ? _____ ans

11. À quel(s) groupe(s) ethnique(s) considérez-vous appartenir? Cochez toutes les réponses qui s'appliquent.

___ Blanc / caucasien (1)
___ Noir (ex., Haïtien, Africain, Jamaïquain) (2)
___ Latino / Hispanique (3)
___ Asiatique (ex., Chinois, Japonais, Vietnamien) (4)
___ Moyen Orient (5)
___ Natif / Première nation / Métis (6)
___ Iles du Pacifique (7)
___ Autre (99), spécifiez : _____

Inventaire Abrégé de Symptômes-18

Version française du *Brief Symptom Inventory-18* (BSI-18) (Derogatis, 2000)

Lisez attentivement chaque item et encerclez le chiffre qui décrit le mieux à quel point ce problème vous a troublé au cours des 7 derniers jours, y compris aujourd'hui.

CE PROBLÈME VOUS A-T-IL TROUBLÉ?	Pas du tout	Un peu	Modérément	Beaucoup	Intensément
1. Évanouissements ou étourdissements	0	1	2	3	4
2. Manque d'intérêt pour tout	0	1	2	3	4
3. Nervosité ou impression de tremblements intérieurs	0	1	2	3	4
4. Douleurs au cœur ou à la poitrine	0	1	2	3	4
5. Sentiment de solitude	0	1	2	3	4
6. Sentiment de tension ou de surexcitation	0	1	2	3	4
7. Nausées ou maux d'estomac	0	1	2	3	4
8. Avoir le cafard	0	1	2	3	4
9. Tendance à vous sentir effrayée sans raison	0	1	2	3	4
10. Difficulté à reprendre votre souffle	0	1	2	3	4
11. Sentiment que vous ne valez rien	0	1	2	3	4
12. Épisodes de terreur ou de panique	0	1	2	3	4
13. Engourdissements ou picotements dans certaines parties du corps	0	1	2	3	4
14. Vous sentir sans espoir face à l'avenir	0	1	2	3	4
15. Vous sentir tellement agitée que vous ne pouvez rester en place	0	1	2	3	4
16. Sentiment de faiblesse au niveau de certaines parties du corps	0	1	2	3	4
17. Pensées d'en finir avec la vie	0	1	2	3	4
18. Sentiment d'avoir peur	0	1	2	3	4

QUESTIONNAIRE SUR LE COUPLE DES PARENTS

1. Quel âge aviez-vous <u>au début</u> de votre relation de couple avec le père de votre enfant survivant de cancer? _____

2. Depuis combien de temps êtes-vous en couple (ou pendant combien de temps avez-vous été en couple) avec le père de votre enfant survivant de cancer? _____

3. Quel est votre statut conjugal actuel ?

   1.___ En couple avec le père de mon enfant survivant de cancer (allez directement à la question 5)

   2.___ En couple avec un autre conjoint (répondez à la question 4)

   3.___ Seule (séparée / divorcée) (répondez à la question 4)

   4.___ Veuve (allez directement à la question 5)

4. Si vous êtes <u>séparée / divorcée du père de votre enfant survivant de cancer</u>, à quel point la maladie de votre enfant a-t-elle contribué à cette séparation selon vous ?

   1.___ La maladie (et le contexte entourant la maladie) n'est *pas du tout liée* à notre séparation.

   2.___ La maladie (et le contexte entourant la maladie) a *légèrement* contribué à notre séparation.

   3.___ La maladie (et le contexte entourant la maladie) a *modérément* contribué à notre séparation.

   4.___ La maladie (et le contexte entourant la maladie) a *beaucoup* contribué à notre séparation.

   5.___ La maladie (et le contexte entourant la maladie) est *entièrement responsable* de notre séparation.

5. Étiez-vous en couple avec le père de votre enfant survivant de cancer <u>au moment où votre enfant était malade et suivi pour des traitements à Sainte-Justine</u>?

   1.___ Oui, nous étions en couple (répondez à la question 6)

   0.___ Non, nous étions séparés (allez directement à la page 15)

Les questions suivantes traitent de la période pendant laquelle votre enfant était malade et suivi pour des traitements à Ste-Justine. SVP répondre à ces questions en repensant à votre relation de couple pendant cette période.

6. Lorsque les couples vivent des situations de stress important, incluant la maladie d'un enfant, il est possible que ces situations influencent leur relation de couple de diverses façons. À l'aide des échelles de réponse suivantes, veuillez évaluer <u>à quel point la maladie de votre enfant a eu un effet</u> sur les dimensions conjugales suivantes :

6.A.1. L'intimité entre vous et votre conjoint :

1	*2*	*3*	*4*	*5*	*6*	*7*
Effet très négatif	*Effet modérément négatif*	*Effet légèrement négatif*	*Aucun effet*	*Effet légèrement positif*	*Effet modérément positif*	*Effet très positif*

6.A.2. Si vous avez répondu 1, 2 ou 3, ces effets négatifs ont-ils perduré lorsque les traitements de votre enfant ont été terminés ?

1.___ Les effets négatifs ont disparu immédiatement
2.___ Les effets négatifs ont perduré, mais se sont estompées avec le temps
3.___ Les effets négatifs ont été permanents

6.B.1. La qualité du soutien entre vous et votre conjoint :

1	*2*	*3*	*4*	*5*	*6*	*7*
Effet très négatif	*Effet modérément négatif*	*Effet légèrement négatif*	*Aucun effet*	*Effet légèrement positif*	*Effet modérément positif*	*Effet très positif*

6.B.2. Si vous avez répondu 1, 2 ou 3, ces effets négatifs ont-ils perduré lorsque les traitements de votre enfant ont été terminés ?

1.___ Les effets négatifs ont disparu immédiatement
2.___ Les effets négatifs ont perduré, mais se sont estompées avec le temps
3.___ Les effets négatifs ont été permanents

6.C.1. La sexualité entre vous et votre conjoint :

1	*2*	*3*	*4*	*5*	*6*	*7*
Effet très négatif	*Effet modérément négatif*	*Effet légèrement négatif*	*Aucun effet*	*Effet légèrement positif*	*Effet modérément positif*	*Effet très positif*

6.C.2. Si vous avez répondu 1, 2 ou 3, ces effets négatifs ont-ils perduré lorsque les traitements de votre enfant ont été terminés ?

1.___ Les effets négatifs ont disparu immédiatement
2.___ Les effets négatifs ont perduré, mais se sont estompées avec le temps
3.___ Les effets négatifs ont été permanents

6.D.1. Les conflits entre vous et votre conjoint :

1	*2*	*3*	*4*	*5*	*6*	*7*
Effet très négatif	*Effet modérément négatif*	*Effet légèrement négatif*	*Aucun effet*	*Effet légèrement positif*	*Effet modérément positif*	*Effet très positif*

6.D.2. Si vous avez répondu 1, 2 ou 3, ces effets négatifs ont-ils perduré lorsque les traitements de votre enfant ont été terminés ?

1.___ Les effets négatifs ont disparu immédiatement
2.___ Les effets négatifs ont perduré, mais se sont estompées avec le temps
3.___ Les effets négatifs ont été permanents

6.E.1. Le temps passé et les activités réalisées avec votre conjoint :

1	*2*	*3*	*4*	*5*	*6*	*7*
Effet très négatif	*Effet modérément négatif*	*Effet légèrement négatif*	*Aucun effet*	*Effet légèrement positif*	*Effet modérément positif*	*Effet très positif*

6.E.2. Si vous avez répondu 1, 2 ou 3, ces effets négatifs ont-ils perduré lorsque les traitements de votre enfant ont été terminés ?

1.___ Les effets négatifs ont disparu immédiatement
2.___ Les effets négatifs ont perduré, mais se sont estompées avec le temps
3.___ Les effets négatifs ont été permanents

6.F.1. Votre satisfaction conjugale générale :

1	*2*	*3*	*4*	*5*	*6*	*7*
Effet très négatif	*Effet modérément négatif*	*Effet légèrement négatif*	*Aucun effet*	*Effet légèrement positif*	*Effet modérément positif*	*Effet très positif*

6.F.2. Si vous avez répondu 1, 2 ou 3, ces effets négatifs ont-ils perduré lorsque les traitements de votre enfant ont été terminés ?

1.___ Les effets négatifs ont disparu immédiatement
2.___ Les effets négatifs ont perduré, mais se sont estompées avec le temps
3.___ Les effets négatifs ont été permanents

7. De façon générale, comment décririez-vous l'impact de la période pendant laquelle votre enfant était malade sur votre relation de couple ?

1	*2*	*3*	*4*	*5*	*6*	*7*
Cette période nous a éloigné / a été néfaste pour notre relation			*Cette période n'a eu aucun effet sur notre relation*			*Cette période nous a rapproché / a renforcé notre relation*

***** Si vous êtes présentement en couple avec le père de votre enfant survivant de cancer, veuillez répondre au questionnaire suivant *****

***** Si vous êtes présentement séparée/divorcée du père de votre enfant survivant de cancer, ne répondez pas au questionnaire suivant et allez directement à la page 15 *****

ÉCHELLE D'AJUSTEMENT DYADIQUE (DAS)

Ce questionnaire porte sur votre perception de votre vie de couple **présentement**. Il s'agit de votre opinion personnelle. Ne soyez pas préoccupée de ce que pourrait répondre votre partenaire. C'est votre avis qui compte ici.

Pour les questions 1 à 3, indiquez votre réponse en encerclant le chiffre approprié.

0 *Toujours*	*1* *La plupart du temps*	*2* *Plus souvent qu'autrement*	*3* *Occasion-nellement*	*4* *Rarement*	*5* *Jamais*

1.	Est-ce qu'il vous arrive souvent ou est-ce qu'il vous est déjà arrivé d'envisager un divorce, une séparation ou de mettre fin à votre relation actuelle?	0	1	2	3	4	5
2.	De façon générale, pouvez-vous dire que les choses vont bien entre vous et votre partenaire?	0	1	2	3	4	5
3	Vous confiez-vous à votre partenaire?	0	1	2	3	4	5

4. Les cases suivantes correspondent à différents degrés de bonheur dans votre relation. La case centrale « heureux » correspond au degré de bonheur retrouvé dans la plupart des relations. Cochez la case qui correspond le mieux au degré de bonheur de votre couple.

☐ 0	☐ 1	☐ 2	☐ 3	☐ 4	☐ 5	☐ 6
Extrêmement Malheureux	Passablement malheureux	Un peu malheureux	Heureux	Très heureux	Extrêmement heureux	Parfaitement heureux